**-DRAFT 8/14/02-**
**HEALTH SERVICES WORKING GROUP**
**STUDY HYPOTHESES FOR THE NATIONAL CHILDREN'S STUDY**

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**Perinatal and Obstetric Services Subcommittee**
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**Executive Summary**

The Health Services Working Group (HSWG) aims to understand the role of the broad spectrum of health services interventions, including health promotion and preventive services, diagnostic, treatment, rehabilitative and supportive services, and health policies, on the physical, psychosocial, and cognitive aspects of health and development across a person's lifespan. There are few nationally representative to show the impact of this spectrum of services on children's health, but the potential for impact is considered to be large and important. Health services have the potential not only to impact on those with special health care needs or those considered at high-risk of developing a condition, but also has a role in promoting optimal health and development in those children without specifically identified problems. Testing the impact of a broad range of services on health and developmental outcomes will require a large-scale longitudinal study.

The HSWG's underlying conceptual approach to the study is a modification of the main model used by the NCS. In this approach, health services interventions can act as a protective modifying or mediating factor in the gene-exposure-outcome relationship in at least 3 distinct parts of the explanatory pathway: 1) health services may actively modify the presence of an exposure; 2)health services can mediate the relationship between being exposed and the development of an adverse health condition; and 3) health services can modify the trajectory of health and developmental outcomes once a condition becomes manifest.

Studying health services in the NCS will raise several methodological issues. First, the study will need to include a set of comprehensive measures of health, functional, and developmental status. This might be done in a stepped approach so that those at least risk or illness or disability receive a basic core set of measures, but those with specific conditions or identified as at-risk for specific conditions receive a more extensive set of measures. Second, measures of health services, including measures of the health systems, measures of specific interventions provided, and measures of quality of the services provided, will need to be included in the study. Third, nested interventional studies should be considered when necessary to test the efficacy of health services when the longitudinal cohort approach is inappropriate for testing hypotheses. Finally, use of innovative sampling techniques such as probability weighting and cluster sampling should be used to allow oversampling of priority populations, but still allow development of a U.S. population representative sample.

Because of the wide range of types of health services and settings for these services, the group has divided into topic-oriented subcommittees, including: mental health services, preventive health services, medical care services, dental health services, community health services, and obstetric and perinatal health services. The group as a whole developed a set of general hypotheses, which serve as guides for the development of specific hypotheses within the topic-oriented areas. The general hypotheses are presented below; specific hypotheses can be found in the body of the document.

H1. Quality health care services can act to mediate the genetic and environmental influences on health and developmental trajectories over a child's lifespan. We define quality health services to have the following dimensions, expanded from the Institute of Medicine criteria: 1) safe; 2)

effective; 3) patient and family centered; 4) timely; 5) efficient; 6) equitable; and 7) coordinated with public health, educational, and social services. Child health includes a focus on promoting optimal health and development, getting better, living with illness, and coping with the end of life. Specifically health services can act as a mediator through the following mechanisms:

a) Through reduction or elimination of adverse environmental exposures

b) Through mediating the relationship between gene-environment interactions and development of health conditions

c) Through mediating the relationship between health conditions and trajectories of health and developmental outcomes.

H2. Quality health care services can reduce disparities in health and developmental outcomes resulting from social, economic, psychologic, and physical environmental disparities.

## Introduction

*The Case for Health Services Research in the NCS*

The goal of the National Children's Study is to study the impact of genetic influences and the physical, social, psychological, and economic environment on the health and development of children. In this planned longitudinal study, the relationships of interest can be viewed using a simplified but traditional epidemiological model of exposure-modifier-outcomes. These exposure-modifier-outcomes relationships can be portrayed as a set of potential etiologic pathways that emerge and develop over the child's lifespan (Figure 1). This model also demonstrates the points at which people, programs, and systems can act to potentially modify a pathway, by introducing a protective factors and risk reduction strategies that modify and mediate the potential outcomes of a particular life path.. Viewing these human interventions as protective factors in a risk-exposure pathway allows us to see clearly how the NCS can contribute to child health, by improving our ability to prevent the onset of disease or abate the course of an illness or injury over the course of the lifespan.

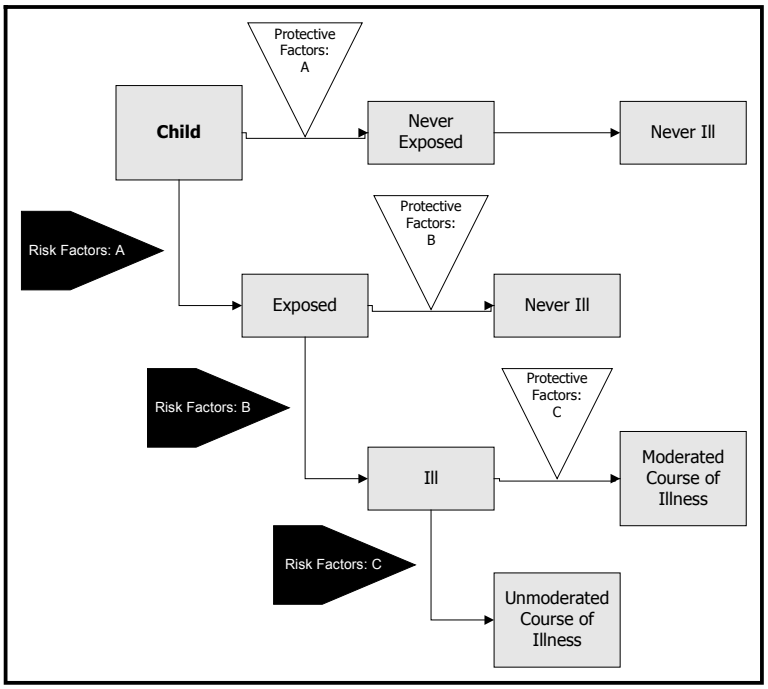



**Figure 1. HSWG conceptual model, top level.**

Using this conceptual approach, the Health Services Working Group (HSWG) aims to understand the role of the broad spectrum of health services interventions, including health promotion and preventive services, diagnostic, treatment, rehabilitative and supportive services, and health policies, on the physical, psychosocial, and cognitive aspects of health and development across a person's lifespan. In this report, we outline our assumptions about the potential impact of health services to improve child health and developmental outcomes based on current epidemiological data, as well as the policy significance that a well developed health

services component of the NCS could contribute to our nations ability to effectively improve the health of all Americans. Building on the conceptual model adopted by the NCS ICC we clarify the roles that health services and measurement of health services impacts can have on the overall design, measurement strategies and potential results of the NCS. We suggest a set of strategies that we believe would allow for the parsimonious measurement of health services impacts on a set of common and representative conditions that we refer to as tracer conditions, and we also suggest selection strategies for prioritizing what conditions and services might be considered for further study. We then present our general framing research questions and exemplar hypothesis before presenting a list of the leading health services hypothesis that emerged from our six subgroups- medical care, mental health services, community health services, preventive and developmental health services, dental health services, and perinatal health services. In the appendix that follows are more detailed reports from each of the subcommittees with detailed presentation of selected hypothesis including the rationales for their selection, and potential implications for study design and measurement.

There are few national representative data to show how much of an impact health services have on children's health, but the unrealized potential for health services to impact on children's health and development is considered to be large and important. Based on current national epidemiological data, we expect approximately 2-4% of children to have severe disabilities due to health conditions (Figure 2). These are children who are limited in their ability to play or go to school due to a severe and chronic disorder or impairment, and traditionally are high and costly users of a wide range of health services. In addition to this small percentage of children with extreme health services needs, , an additional 10-16% of children will have ongoing special health care needs, as defined by the Bureau of Maternal Child Health definition and recent national surveys. These are also a group of children that are high utilizers of health care services with a range of different and often time limited conditions and impairments. While much more difficult to specify there are another 30-40% children who may have or be at significant risk for behavioral, mental health or learning problems based on data from NCHS and other more localized epidemiological, educational and service delivery studies. This is an important an as of yet not well described and studied population with a range of developmental risks, and full fledged diagnosis that have not been well studied. This large group of children who are not functioning up to their full potential represent a population of significant importance because of the potential burdens they place on other systems (e.g education, juvenile justice, social service) and because a developing literature indicate that the potential to intervene and modify developmental health trajectories is actually quite large. Even the 60-70% not identified as having a specific health or developmental problem could be impacted in a positive way by the health care system, optimizing developmental and functional outcomes, adding years of healthy life, as a result of health promotion and preventive services.

The HSWG adopted a definition of health consistent with the Ottawa Charter (1986) that we adapted to focus on the particular health concerns of children- *Health is the extent to which a child or a population or group of children is able to realize their own aspirations and the aspirations of their parents and society, to satisfy needs, and to change, cope and adapt to the environment in which they live. Health is therefore a resource for everyday life, not the objective*

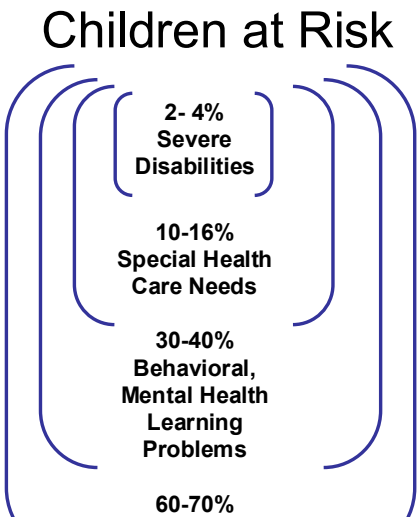



**Figure 2. Children at risk for health conditions**

*of living; it is a positive developmental concept, emphasizing the development of social and personal resources and well as the development of physical capacities* For the majority of children not identified as having a diagnosable condition, we need to know not only how much of an impact health care has on reaching optimal health and development, but how to optimize health services to reach these ideals.

Attaining these goals of optimal health and development implies not only treatment of identified conditions, but promotion of health to enable children and their families to increase control over and to improve their health (Ottawa Charter, 1986). These activities can occur in a variety of venues. Therefore health promotion goes well beyond the responsibility of health care services as traditionally considered to be composed of clinical services administered in outpatient and in-patient settings. Accordingly, we have taken a broad definition of what comprises children's health services for the purpose of this study, including traditional acute and chronic medical care services, mental health services, perinatal services, dental health services, preventive and anticipatory guidance services, and community health services and policy.

We considered the importance of understanding the impact of health services on children's health and development from several perspectives

- The cost of health care for children and the potential of averting future costs if health and development is optimized at the earliest point possible
- The potential under investment in children's health care relative to the current expenditures for adults, especially at the end of the life
- The importance of having new, representative data on the missed opportunities to improve the health of children, to maximize their life long learning potential, and to enhance their long term contribution to shared social goals.

In the year 2000, the United States spent $1.3 trillion dollars, over 13% of the GDP, on health care (Stat Abstract, 2001) to improve health over the lifespan, yet there are surprisingly few data on the impact of these large expenditures on improving health outcomes. Children are an especially important group to study, given what is increasingly known about the long reach of the early years on adult health and social outcomes. Several recent studies have pointed to the role of pre-disease pathways that have their origins in critical and sensitive developmental periods early in life that influence health and development across the lifespan. Irrespective of the potential gain the Federal Government spends much less per capita for health and other services on children than on other segments of the population. For example, in 2000 federal health spending on children under age 18 was estimated at $25 billion on Medicaid and the State Children's Health Insurance Program (SCHIP) and $123 billion on other services, including food

stamps, nutrition programs, social services, and other health and human development programs(CBO, 2000). Decision makers need to know whether these expenditures improve the health of children and whether further expenditures for specific programs aimed at children could further improve health and development. Not only is the study of the impact of these services on health across the lifespan important in its own right, but from a policy standpoint, stakeholders need to know the benefit of these expenditures on health services in influencing health and development when considering tradeoffs between different health programs. Moreover, if our country wants to entertain new policies that would enhance child health promotion and disease prevention efforts, there will be an important need for evidence of the potential long term impacts on children as they develop into adults, as well as the potential benefits to society. The NCS has the unique potential to provide exactly this kind of information.

The study of health services will be a necessary consideration even for those working groups not directly concerned with the impact of health services on outcomes. Most NCS working groups have adopted a framework that generally examines the impact of environmental exposures (broadly defined) on relevant outcomes. While we discuss our conceptual framework in detail below, an underlying assumption of our approach is that health services can act to reduce exposures or as a mediator in the gene-environment-outcome relationship. This is a critical point for two reasons. First, use of health services will need to be measured and controlled for to accurately understand the gene-environment-outcome relationship, even when use of health services are not the principle focus of the primary research question, because health services can alter the outcomes resulting from the exposure of interest. The second reason deals with health disparities. To the extent that disparities in exposure are seen in the study, e.g. socioeconomic disparities and disparities in environmental exposures, the use of effective health care services may act to improve health through reducing the disparities in health and development outcomes that might otherwise have occurred from the disparities in exposures.

*Conceptual Framework for the Health Services Working Group*

The NCS Interagency Coordinating Committee (ICC) has developed a basic conceptual model to organize data collection and to specify the relationship between health outcomes and number of different explanatory factors. These factors include social, environmental, infectious and chemical exposures and other influences that contribute to or are somehow associated with the outcomes of interest. Figure 3 is the graphic depiction of the proposed model.
The conceptual model that the ICC has adopted also suggests that there are mediating factors, which can modify the effect and impact of different exposures on the relevant health outcome.

*The Role of Health Services*

Beginning with this conceptual approach, the NCS Health Services Working Group (HSWG) has maintained the basic model but has highlighted the critical role that health services play as mediators in this causal pathway (Figure 1). In the modified figure the HSWG suggests that health services can act as a protective factor for developing disease or disability, and play a crucial mediating role in at least 3 distinct parts of this proposed explanatory pathway (Protective factor A, Figure 1). The first is the role that health services might play to actively modify the presence of an exposure. For example, specific public policies may reduce a child's or teen's access to alcohol and resulting alcohol abuse and alcohol-related injuries, and firearm-related injuries may be prevented by clinician counseling of parents in firearm safety to prevent children's access.

Secondly, health services can directly mediate the causal pathway by modifying the relationship between exposure and the onset of a disease process, once the individual is exposed, and altering the nature of the exposure-outcome path (Protective factor B, Figure 1). For example, a school-based program promoting physical activity may reduce the probability of obesity in children who are genetically predisposed or in those without physically active role models in their social environment.

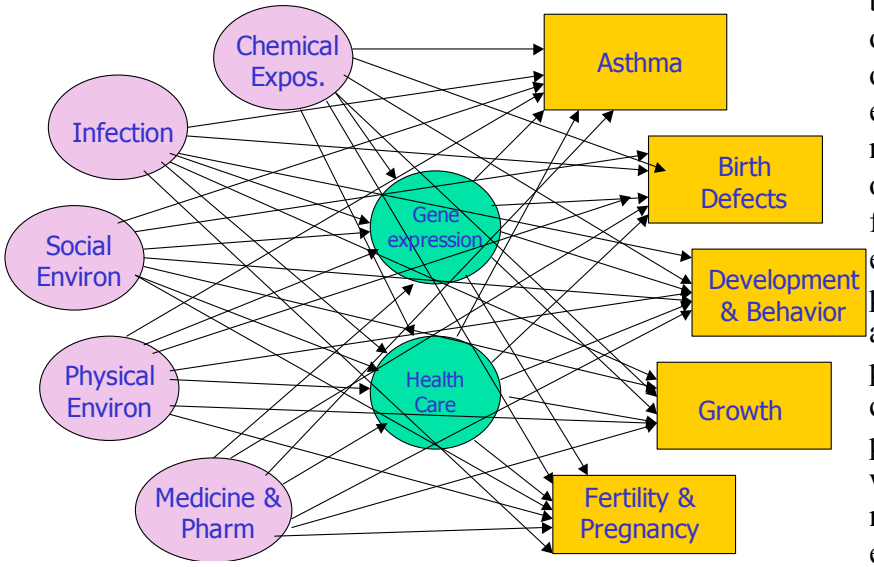



**Figure 3. NCS Study Model**

The third way that health services can intervene to change the course of this pathway is once an outcome begins to manifest (Protective factor C, Figure 1). This is through the role that health services play in reduction or modification of the actual disease burden or altering the rehabilitation or habilitation process. For example, a well-coordinated asthma care system including primary and subspecialty clinicians, home-based services, and school-based services may modify asthma severity and recurrences, reduce the functional disability due to asthma, and mediate the trajectory of the disease process directly as a result of early identification of exacerbations and early initiation of effective therapies.

Different aspects of the health care system might provide each of these distinct and critical mediating roles of health services. For example many primary preventive services that reduce the presence of exposures are provided through community and public health service systems, e.g. the "Back to Sleep" mass media campaign for reduction of sudden infant death syndrome (SIDS) could be considered a public health service which occurred outside of the traditional clinical settings. The interventions that target the relationship between exposure and manifestation of the disease pathway is provided by traditional preventive, diagnostic and treatment services. The

third point of intervention is part of on-going medical care and chronic disease management that determines the shape and trajectory during this rehabilitative period.

We wish to emphasize that each of these potential points of health service mediation can have significant impact on health and functional outcomes. These services have the potential to modify the trajectory of health in an individual or in the population as a whole. The impact of these services may not only vary across individual children and across different environmental exposures, but they may also vary across intrinsic factors of the health care delivery system; for example, use of specific preventive services might vary by geographic locale and by whether or not the child is enrolled in a managed care organization.

Figure 4 depicts the working group's conceptualization of broadly classifying the study of health services across a variety of health care systems and settings. In this conceptual framework, a service can be placed in a grid with three axes: focus of the service, components of quality of the service, and whether or not the service is applicable to specific tracer conditions. Specific hypotheses may address one or more points within this grid. Focus of the services, which represents a modification of the Foundation for Accountability (FACCT) model that was adopted by the IOM in its Crossing the Quality Chasm report, include services that promote optimal health and development and prevent illness, getting better, living with chronic illness, and coping with the end of life. The second axis represents the working group's extension of the Institute of Medicine classification of essential elements that characterizes good quality health care within a broad conceptual framework of children's health services. These attributes are particular applicable to each of the three example health services mediation points in the diagram. They include: safety, effectiveness, patient- and family-centeredness, equity, and co-ordination with other social services. The use of these system attributes can be exemplified in the newborn whose mother has HIV and whose own exposure might be modified by prenatal provision of antiretroviral drugs and counseling about breastfeeding. In order for this health service pathway to be analyzed it is important to consider components of the health system such as access to prenatal care, appropriateness of the services provided, or coordination with social services to ensure adequate financial resources to obtain therapy and to ensure proper follow-up.

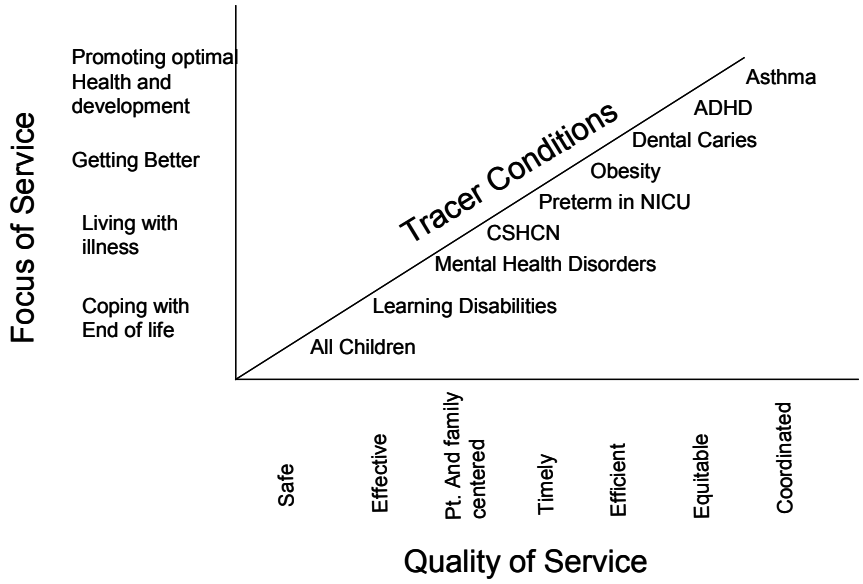



**Figure 4. Conceptualization of specific health services.**

Finally, the third axis in the diagram represents the population to be studied for a particular hypothesis. Some services are targeted to all children, such as common preventive services. Some services are only applicable to children with specific conditions. For these services, the working group has chosen to recommend studying specific tracer conditions.

## Methodological and Strategic Considerations

*Measurement*

One of the most important methodological contributions that health services has made over the past decade is better specification in the measurement of health conditions, quality of life, child well being, and developmental potential as well as new integrated measures of health related to quality of life and disability-adjusted life years. Each of these different types and dimensions of health measurement has been a major focus of instrument development and measurement design in health services field. In addition, because the health services field has been particularly interested in partitioning the effect of health services and specifying the unique contribution of health services in relationship to other determinants of health, health services research has developed methods and analytic techniques for analyzing the contributions of individuals and family behaviors, personal and family resources, working conditions and other roles of interactions of physical, social, cultural and policy contacts on health outcomes.

Also important to the study of health services is measurement of the content and quality of health services. Measures of the structure and performance of the health care system have been developed for specific situations, although additional measures will likely need developed for the broad range of conditions potentially examined in the NCS. Since quality of services provided will be a key mediating factor of health outcomes, measures of the content of health services and the quality of these services will need to be included in the study. Many of these measures will need to be obtained from the health care system, e.g. through medical record review or through health system administrative data. Some measures of content of care may be obtainable through parental report, and the working group finds that a pilot study should be commissioned to validate these measures against medical record data or administrative data on content of care.

The HSWG finds that the NCS will need to include a comprehensive set of measures of health, functional, and developmental status in the study. The group recommends that a basic core set of measures be developed for all children in the study, and that additional measures be included for specific children within the study. In parallel, we recommend that basic information on health services provided be collected for all children. For those children with specific conditions, functional deficits, or developmental disabilities, more detailed information on the health services available and those utilized will need to be collected. For the approximately 60% of the child population that at any point in time is in "good enough" health with the major potential impact of health services having to do with their level of developmental function or flourishing. For this group of reasonably healthy children, the measures of health and developmental function are likely to be relatively stable and relatively uneventful throughout the course of the entire study. This will include children who are never exposed or have low levels of exposures with immeasurable impacts, and who are rarely ill. Nonetheless, the role health services could

conceivably mediate their overall developmental potential and level of flourishing and influence their long term health and developmental trajectories, particularly in the case of low income or otherwise disadvantaged children, whose overall capacity to respond to additional and even mild adversity may be limited.

The second group of children, are those who are at risk due to documented and specific exposure that are measured at some point during the study, or children who develop a medical or mental health condition. These children, where exposure(s) can be documented, may avoid illness either because the role that health service plays in modulating exposure or because of their individual genetic makeup and response patterns and other individual variations in how the exposure is mediated.

A third group of children are likely to get ill as a result of measurable exposure or other potentially documentable factors. For that group of children, health services may modify the course of illness and the impact of health services can be measured in terms of different functional outcomes, quality of life, and cost related issues. For those children who do develop a condition- from Cerebral Palsy to Acute Lymphoblastic Leukemia it will be important to measure the mediating role of the content, availability, quality and continuity of health care on their long-term outcomes.

In parallel to the study of the impact of health services on outcomes for the study cohort as a whole, the HSWG also recommends the development of set of tracer conditions that can be used to examine the role of health services, in its primary, secondary and tertiary roles. These tracer health and developmental conditions are common and reflect different types of condition categories and different temporal patterns of expression across the life course. This includes conditions that are chronic and persistent, chronic and relapsing, and acute with variable time courses. We have also selected conditions that may be important at different ages and developmental stages, e.g. learning disabilities may not be evident until approximately age 5, whereas language impairment is evident in the preschool years. The tracer conditions that are suggested for consideration include:

- Asthma
- ADHD
- Mental Health disorders
- Language impairment
- Mental retardation (mild to moderate)
- Children with special health care needs (in 3-4 subcategories including CP, Congenital Heart Disease, Chronic Lung disease)
- Preterm infants graduating from NICUs.
- Obesity
- Dental Caries
- Learning Disabilities

By a more focused assessment of the natural history and role of health care on each of these specific tracers conditions, it will allow for a more specific focus on some of the key research hypotheses for the overall study. The HSWG has developed specific hypotheses for some,

although not all of the suggested tracer conditions. As interest in specific areas and conditions develop in the NCS as a whole, representative hypotheses could easily be developed for any and all of these tracer conditions.

In considering what and how to measure the impact of health services on health outcomes the HSWG is struck by a basic tension in how health care services are delivered and how health care impacts are measured. Throughout the US there are a large number of health services that we already know are not having much impact because they are being provided at an insufficient level or with such poor quality as to have virtually no real effect. Therefore in measuring what is done it will also be necessary to measure the quality of how those services are provided and received. Many of these measures will need to be developed and piloted for this study.

The HSWG has also noted that many of our currently available measures of health status, functional status and quality of life, while potentially adequate for basic measures, are not all that they could and should be. Therefore the HSWG has suggested a 2-tier measurement strategy.

First tier – Focus is on what is currently provided as part of routine care without any special intervention. Measure these impacts using what is currently state of the art measurement.

Second tier – Includes the development of better measurement capacity including better measures of quality of children's health services (e.g. measures of co-ordination of care); better measures of developmental function and its relationship with disease; better measures of quality of life; and better measures of physical activity and other health and functional attributes that are important for individual and population health measurement.

*Selection of Conditions and Exposures to Study*

I. Focus investigation on target conditions, exposures, or interventions that are likely to be 'important'.

Conditions or exposures can be important because they are highly prevalent, or have a high impact on health, or both. This criteria suggests a system of prioritizing study questions based on the degree of prevalence or incidence and the impact of health, with high degrees for both attributes given the greatest weight (+++):

| | | **Impact on Health** | | |
|---|---|---|---|---|
| | | Low | Medium | High |
| **Prevalence or Incidence of Condition or Exposure** | Low | | | + |
| | Medium | | + | ++ |
| | High | + | ++ | +++ |

Health service interventions can be important because many children are exposed to them (that is, they are quite prevalent or frequent), or because they have a high impact on the health of the recipient, or both. Just as above, this suggests a scheme for prioritizing:

| | | **Impact on Health** | | |
|---|---|---|---|---|
| | | Low | Medium | High |
| **Prevalence or Incidence of Intervention** | Low | | | + |
| | Medium | | + | ++ |
| | High | + | ++ | +++ |

A third attribute needs also to be considered. Certain other conditions, exposures, or interventions assume a higher priority due to disparities across groups of people in our society, such as the disparate exposure to lead or the unequal access to certain beneficial interventions, such as immunization. In such cases, the scheme detects important study questions by detecting discrepancies in the prevalence or incidence across groups, or disparities in the impact on health. In the table below, both Groups A and B experience a high level of exposure or condition, but the health impact of this exposure or condition differs substantially. Groups A and B could be defined by socioeconomic characteristics, genetic constitution, or whether or not members received a specific health service. Such a situation is ideally suited to the NCS to determine the reasons for the discrepant health impacts.

| | | **Impact on Health** | | | | | |
|---|---|---|---|---|---|---|---|
| | | *Group A* | | | *Group B* | | |
| | | Low | Medium | High | Low | Medium | High |
| **Prevalence or Incidence of Condition or Exposure** | Low | | | | | | |
| | Medium | | | | | | |
| | High | A | | | | | B |

In the table below, Group A has a low level of exposure to a health care intervention while group B has a high level of exposure, with high impact on health for both groups. Such a scenario would be worthy of study by the NCS in part to determine reasons for the disparity of health care exposure and in part of reasons of study power (see next section).

| | | **Impact on Health** | | | | | |
|---|---|---|---|---|---|---|---|
| | | *Group A* | | | *Group B* | | |
| | | Low | Medium | High | Low | Medium | High |
| **Prevalence or Incidence of Intervention** | Low | | | A | | | |
| | Medium | | | | | | |
| | High | | | | | | B |

II. Focus evaluation on exposure-modifier-outcome relationships that require large cohort.

Power calculations will need to account for the investigators' intention to examine not just exposure-outcome relationships, but also how these relationships are modified by other exposures (such as secondary or tertiary preventive health care), or by individuals' socioeconomic characteristics (that is, when examining racial disparities, across SES strata). The power of these analyses will be greatly influenced by the analytic cells with the smallest number

of subjects. Accordingly, even though some of the exposures are quite prevalence and the outcomes are not rare, investigation of effect modification and sub-group analyses will require a considerable number of subjects.

III. Focus evaluation on health service interventions that are very unlikely to be studied through randomized controlled trials.

The impact of a health service intervention is best studied in randomized controlled trials. Nevertheless, in reality there are a host of reasons why certain interventions are highly unlikely to be studied using this gold-standard methodology. The observational aspects of the NCS health services research component should therefore focus on interventions that are likely to be important but are even more likely to never be evaluated as rigorously as the NCS could do.

*Interventional Studies in the NCS*

While the HSWG believes that there are health service interventions for which it is infeasible to test in interventional studies as above, we emphasize that there are opportunities for intervention. We find that it is advisable to consider ways the National Children's Study can introduce plan variation and nesting of specific interventions in order to take advantage of this large sample size and opportunity to test the effectiveness and impact of specific interventions. This approach would be similar to other major longitudinal studies conducted in other countries, as well as the US Women's Health Initiative. For example, in the ALSPAC longitudinal study of children in Bristol, England a nested intervention study determined the benefits of amblyopia screening (4) in the pediatric population. The NCS provides enormous opportunities to test interventions to determine not only the efficacy of interventions but also to look at the effectiveness of interventions where only small-scale trials have thus far been completed. For health service interventions that have been proven effective, the study could consider quality improvement trials. These trials test the effectiveness of alternative strategies to implement interventions into everyday healthcare delivery settings.

*Sampling Strategies*

The HSWG believes that this large-scale study of the health of children should be generalizable to the U.S. population of children. We recognize that over-sampling based upon specific demographic factors or exposures will be desirable and necessary for the conduct of the study. However, we also recognize that for some research questions, a representative sample will be necessary. In addition, having a representative sample will provide sufficient variability of dimensions as of yet currently unknown, but may become important as new knowledge develops as the study proceeds; thus, selection of a representative sample enhance the ability to answer questions during the full course of the study, not just the ones we know to ask now. Finally, children drawn from easily recruited sites are likely to differ in critical and difficult to measure ways from children recruited at random. If any of these differences interact with important analytic concerns, the opportunity for bias arises. For these reasons, the HSWG recommends use of statistical techniques such as probability weighting and cluster sampling so that a U.S. representative sample can be developed from the cohort recruited into the study.

**General Hypotheses for the NCS Health Services Working Group**

In considering the overall goals of the NCS, the HSWG recognizes the enormous opportunities to address important research and policy questions. These include health services research questions that will be essential to the overall goals of the study, as well as questions whose answers are aimed at improving practice and policy in the U.S. health care system.

The HSWG developed general hypotheses based upon our conceptual approach outlines above. These general hypotheses should be considered as the HSWG's guiding meta-hypotheses that informed the development of the specific hypotheses that are listed in the next section and more fully presented in the appendix. It would be infeasible to test the general hypotheses for all services for all children, even in a study as large as the NCS, since the hypotheses cover such broad areas of health services research. Specific components of these hypotheses, however, can be tested in the NCS. For example, improvement of health outcomes through reduction of exposure can be tested for programs aimed at reducing exposure to tobacco smoke, with their subsequent impact of development and severity of asthma. In this manner, our general hypotheses represent the overarching goals of our research recommendations, and are used to formulate the specific objectives as represented by our specific hypotheses in specific areas of health services. In the following hypotheses, we define the environment broadly to include the social, economic, psychologic, and physical environment.

We present the general HSWG hypotheses below in italics. For each general hypothesis, we present exemplar specific hypotheses, developed by our subcommittees, aimed at testing one specific area of the general hypothesis.

*H1. Quality health care services can act to mediate the genetic and environmental influences on health and developmental trajectories over a child's lifespan. We define quality health services to have the following dimensions, expanded from the Institute of Medicine criteria: 1) safe; 2) effective; 3) patient and family centered; 4) timely; 5) efficient; 6) equitable; and 7) coordinated with public health, educational, and social services. Child health includes a focus on promoting optimal health and development, getting better, living with illness, and coping with the end of life. Specifically health services can act as a mediator through the following mechanisms:*

*a) Through reduction or elimination of adverse environmental exposures*

States with increased alcohol taxes AND with increased allocation of those revenues to adolescent alcohol abuse prevention and treatment programs will have the lowest rates adolescent alcohol misuse, suggesting a generalizable model of state-level policy and adolescent health-related behaviors (Community Health).

Health services used by one generation affects the birth outcomes, health service utilization, and childhood health outcomes of the next generation (Perinatal/Obstetric)

*b) Through mediating the relationship between gene-environment interactions and development of health conditions*

Behavioral and developmental screening is optimized when critical and sensitive time periods are identified (Preventive Services).

Children who attend child care centers and schools that have comprehensive, enjoyable programs that promote physical activity and active lifestyles will have lower BMIs and a higher proportion of children that exercise when compared to children attending centers or schools without such programs (Medical Care).

*c) Through mediating the relationship between health conditions and trajectories of health and developmental outcomes.*

CSHCN who have a medical home are more likely to receive comprehensive preventive and acute health care. CSHCN with a medical home have improved health outcomes and the care provided is more cost effective. Sub-hypotheses to be tested include:
-Health care systems that employ electronic medical records will better coordinate care for CSHCN.
-Children in health care systems with defined relationships with schools and other community resources will be more likely to have a medical home and improved outcomes. (Medical Care)

Children with cleft lip and/or cleft palate who are treated by craniofacial teams have better health outcomes than children not treated by craniofacial teams (Dental Health).

*H2. Quality health care services can reduce disparities in health and developmental outcomes resulting from social, economic, psychologic, and physical environmental disparities.*

Communities with enriched early family intervention programs, those with high levels of social capital and those with supplemented child care programs will have better outcomes for infants and toddlers with depressed mothers (Mental Health).

**List of Specific Hypotheses:**

Children's health services research represents a broad field of study that draws upon numerous areas of expertise from clinical, biologic, social science, psychologic, epidemiologic, economic, and statistical fields. The HSWG represents a multidisciplinary expert group with a broad range of expertise. In order to help focus the specific hypotheses for this working group, we have chosen to form six subcommittees representing common and important areas of health services delivery. These subcommittees include the mental health, preventive services, medical care, dental care, community health services, and perinatal areas. The following sets of hypotheses represent the priority hypotheses in each of these areas, as identified by our subcommittees. We include here solely a listing of the hypotheses, but include in the Appendix the complete reports of the subcommittees. These hypotheses are illustrative hypotheses that could inform the Advisory Committee in their deliberations; we do not presume to provide an exhaustive list of deserving health services related hypotheses that could be tested over the entire course of the study.

*Mental Health Services*

MH-1a) Early identification and evidence-based treatment of maternal depression will be related to better infant and toddler functioning (age-appropriate learning and problem solving, communication, mobility, self-care and socialization).

MH-1b) Communities with enriched early family intervention programs, those with high levels of social capital and those with supplemented child care programs will have better outcomes for infants and toddlers with depressed mothers.

MH-2a) Early identification in the disease process for identified mental health issues (those with efficacious treatments) is more likely to lead to treatment.

MH-2b) Early identification will vary by family, child, practitioner, health care system and community characteristics, including post trauma responses to bioterrorism and other disasters.

MH-2c) Early assessment of parental attitudes, ability to act as child advocates and willingness to seek help would identify the most important early opportunities to make families better prepared to seek and obtain treatment.

MH-3a) Evidence-based care for key identified mental problems delivered within routine clinical practice will improve child functioning.

MH-3b) Receipt of evidence-based care will vary by out-of-pocket costs, parental attitudes and community characteristics.

MH-4) Organization and financing features of children's primary health care and mental health services are related to access, quality and cost.

*Prevention*

PS-1a) Expanded, mandatory newborn screening (e.g,. screening for CF and other conditions soon to be identified through human genome project) does not improve health of children.

PS-1b) Behavioral and developmental testing is optimized when critical/sensitive time periods are identified.

PS-1c) Selected screenings for targeted populations are highly effective.

PS-1d) Universal screenings for selected conditions are highly effective.

PS-2a) Office based anticipatory guidance is most effective when broad based community approaches are used to improve health related behaviors.

PS-2b) Office based anticipatory guidance has varying effectiveness for subsets of the population at critical/sensitive time periods.

*Medical Care*

MC-1) The proportion of children who have up-to-date immunization status will be greater in health care systems that utilize electronic medical records (EMR). Sub-hypotheses include:

- Effect of EMR will be greater for newly recommended compared to established immunizations.
- Effect of EMR will be greater for historically-under-immunized groups of children.

MC-2) Genotypic differences across individuals modify the risk of post-immunization known and hypothesized adverse events, including seizures and the development of autism.

MC-3) The degree of benefit of risk information obtained from genomic information for a child will be a dependent on the availability of preventive and/or therapeutic options to manage genetic risk and the degree to which the child's providers have expertise in the use of genomic information. This information may overall be harmful to the extent that the information increases child and parental anxiety, health care utilization, and health care costs in the absence of well-defined and well-communicated risk management options. The overall benefit of this genomic information will be modified by:

- The child's age
- The patient-centered-ness of medical care
- The immediacy of perceived risk
- The efficacy of available interventions against the target disorder

MC-4) Following the death of a child, siblings and parents who receive bereavement care have better outcomes than those who do not. Sub-hypotheses include:

- Impact of bereavement care for siblings will vary by the sibling's developmental age.
- Impact of bereavement care for parents will vary by the age of the deceased child and the manner of that child's death (acutely traumatic versus terminal chronic illness).

MC-5) Surgical outcomes are superior for children who are cared for in children's hospitals than children cared for in community hospitals.

MC-6) CSHCN who have a medical home are more likely to receive comprehensive preventive and acute health care. CSHCN with a medical home have improved health outcomes and the care provided is more cost effective. Sub-hypotheses to be tested include:

- Health care systems that employ electronic medical records will better coordinate care for CSHCN.
- Children in health care systems with defined relationships with schools and other community resources will be more likely to have a medical home and improved outcomes.

MC-7) Children who have a Medical Home are more likely to receive comprehensive, quality health care that achieves cost-effective health care and improved health outcomes for the child and family. A sub-hypothesis:

- Children in health care systems with defined relationships with schools and other community resources will be more likely to have a medical home and improved outcomes.

MC-8) Children who attend child care centers and schools that have comprehensive, enjoyable programs that promote physical activity and active lifestyles will have lower BMIs and a higher proportion of children that exercise when compared to children attending centers or schools without such programs.

*Dental Health Services*

DH-1a) Children's risk of developing dental caries is inversely related to the degree to which they receive recommended well-child services from primary medical care providers.

DH-1b) Children's utilization of dental services is positively related to the degree to which they receive recommended well-child services from primary medical care providers.

DH-2a) Children's risk of developing dental caries is inversely related to the degree to which they receive early and periodic oral health services from dental care providers.

DH-2b) Children who receive early and regular oral health services from dental care providers have less untreated dental disease, lower overall dental and health care costs, and improved oral health-related quality of life compared to children who do not receive early and regular dental services.

DH-3a) Children covered by medical and/or dental insurance are more likely to receive dental services.

DH-3b) The quality of children's medical and/or dental insurance is related to the extent to which they receive dental services.

DH-3c) Children whose parents have medical and/or dental insurance coverage have increased access to and use of oral health services compared to children whose parents lack medical and/or dental insurance coverage.

DH-4a) The timing and quality of health services for cleft lip and/or cleft palate affects outcomes.

DH-4b) Children with cleft lip and/or cleft palate who are treated by craniofacial teams have better health outcomes than children not treated by craniofacial teams.

*Community Health Services*

CH-1) Initiation of tobacco smoking during adolescence exhibits a high correlation with smoking rates during adulthood and, thereby, with the long-term consequences of smoking. Factors that reduce initiation of smoking during teenage years may, therefore, have great effects on modifying the disastrous consequences of tobacco smoking on the health of the public. Such factors include actions taken by state governments, most prominently through modifying the cost of cigarettes through taxation. The extent of that taxation *and* the programs put in place via use of funds derived from such taxes should be demonstrably correlated with rates of initiation of smoking.

States with increased tobacco taxes AND with increased allocation of those revenues to adolescent smoking prevention and treatment programs will have the lowest rates of adolescent smoking initiation and addiction, after controlling for individual level covariates.

CH-1a) Social disparities in adolescent smoking rates will initially increase as the beneficial effects of state level policy on tobacco use first influence more socioeconomically advantaged youth, before diffusing to less advantaged youth.

CH-1b) States with increased alcohol taxes AND with increased allocation of those revenues to adolescent alcohol abuse prevention and treatment programs will have the lowest rates adolescent alcohol misuse, suggesting a generalizable model of state-level policy and adolescent health-related behaviors.

CH-2) The nature and comprehensiveness of health services provided in settings outside traditional provider/institutional locations may exert major effects on the impact of chronic disease.

Children with asthma who attend schools with effective school-based health services, will have improved respiratory function, fewer acute health care visits and fewer asthma-related school absences, after controlling for sociodemographic and primary health care related variables.

CH-2a) School-based health services that include communication between school nurse and primary care providers, on site asthma management plans, and the capacity for direct administration of daily medicines and peak flow monitoring will be associated with the largest reductions in child morbidity.

CH-2b) Socioeconomically advantaged children with asthma will be more likely to attend schools with effective health services, while socioeconomically disadvantaged children will be more likely to demonstrate benefits from effective school health services.

CH-2c) Primary care services that articulate with school health services will lead to improved asthma outcomes.

CH-2d) Children with ADHD who attend schools with effective school-based health services, will have improved classroom behavior, fewer school disciplinary actions, and fewer school absences, suggesting a generalizable framework relating school health services and health outcomes for children with chronic conditions.

CH-3) Successful school performance and learning represents an outcome of central importance in the well-being of children and youth. Retention in grade, during early years of school, correlates directly with unsuccessful outcomes during later years of school. The capacity of children to enter the educational system effectively may be modified or enhanced if community-based programs are available for health and development promotion and early intervention of at-risk children prior to entering school. The nature and extent of *primary prevention and early identification/intervention programs* within communities becomes potentially a variable of great significance in relation to overall school outcomes.

Increased availability, quality, coordination and outreach of infancy and early child development programs will reduce the incidence of grade retention during the elementary school years, with greatest reductions evident in Grade 1.

CH-4) Neighborhood factors that promote healthy diets and physical activity reduce the likelihood of obesity.

CH-4a) Community violence, exposure to community violence and parental fear of community violence are all associated with decreased outdoor physical activity and increased likelihood of obesity.

CH-4b) Poor school-based nutrition, including non-nutritious school lunch and vending machines, and reduced school-based activity is associated with an increased likelihood of obesity.

*Perinatal and Obstetric Services*

OB-1) Health services used by one generation affects the birth outcomes, health service utilization, and childhood health outcomes of the next generation.

OB-2a) Greater utilization of prenatal care will result in greater utilization of well-child care, sick-child care, and dental care.

OB-2b)Greater utilization of prenatal care and subsequent health services utilization will affect subsequent health behaviors, including preventive health behaviors, breastfeeding and seatbelt usage.

OB-2c) Through utilization of these services, long-term outcomes will be improved.

OB-3a) Content of prenatal care will have a positive impact on short- and long-term childhood outcomes when that care is safe, effective, patient- and family-centered, timely, efficient, equitable and coordinated with public health, educational, and social services.

OB-3b) Content of prenatal care will have a negative impact or will have a less than maximum positive impact when that care is lacking in one of the characteristics listed above.

OB-3c) Content of prenatal care will have influence the clinical decisions that are made during pregnancy, delivery and perinatal care. These decisions will impact short- and long-term childhood outcomes.

**Implications of Conceptual Approach and Working Group Hypotheses for NCS Pilot Studies**

Members of the HSWG have identified the need for specific pilot studies in relevant to incorporation of health services research into the NCS. The following topics have been identified as priorities for white papers, workshops, and pilot studies.

*Commissioned Papers*

- Review of lessons learned from other large national prospective studies, including opportunities for nesting intervention trials in observational studies.
-Evidence for content of prenatal care services.
- Use of decision modeling based upon available data to project outcomes for sample size estimation.
- Optimal periodicity of child assessments as a function of age, developmental stage, and health status.
- Development of nested intervention studies in a longitudinal cohort study of children.

*Workshops*

- Cluster sampling and the implications for analyses.
- Measures of health status, development, function, and severity of illness which can be used across the lifespan and measures which can be used across specific developmental stages (will also need pilot studies for development and testing of measures).
- Measures of health care service structure, content, and quality.

*Pilot Studies*

- Validity of parental (and child) report of health services against medical record review or administrative data.
- Validity of retrospective reports and data analyses of parental preconception factors.
- Development and validation of measures of continuity and of coordination of care.
- Level of respondent burden of measures in different groups.

**APPENDIX:**
**Reports of Topic-Specific Subcommittees Including Hypotheses and Rationale**
**Mental Health Services**
**Preventive Services**
**Medical Care Services**
**Dental Health Services**
**Community Health Services**
**Perinatal and Obstetric Services**

## Mental Health Services Hypotheses for the National Children's Study

**Background Rationale:**

- 20-25% of US children have a diagnosable behavioral/ emotional problem or enough symptoms to place them above a threshold indicating the need for intervention. This amounts to 7-8 million children/ adolescents in any one year. Worldwide mental health issues are on the rise and cause considerable burden to youth and their families (Surgeon General's Report on Mental Health, 2000; Blueprint for Change: Research on Child and Adolescent Mental Health, 2001).

- Few of the children/ adolescents with significant mental health issues receive services. This unmet need continues in spite of the development of validated treatments and service delivery mandates. There are few data on why youth do and do not receive services. There is also little treatment of a well-recognized antecedent of children's mental health problems, maternal depression (Blueprint for Change, 2001; Behavioral Health Management, 2002).

    - As pointed out in NIMH's Blueprint Report: A child's environment, both in and out of the womb, plays a large role in shaping brain development and subsequent behavior. Studies of the care giving environment suggest that extreme environments (such as abuse and neglect) may affect brain cell survival, neuron density, and neurochemical aspects of brain development, as well as behavioral reactivity to stress in childhood and adulthood". Mental health services may be the only avenue to look at some of these extreme environments (identification) and mediate or moderate their effect.

- Little is known about the services children receive. The studies available indicate that state-of-the-art evidence based therapies are not available in routine care (Blueprint for Change, 2001).

- Virtually nothing is known about the impact of organizational and financing issues on the use of mental health services.

- Mental Health, a Report of the Surgeon General (1999), Report of the Surgeon General's Conference on Children's Mental Health and Healthy People 2010 all stress the importance of early identification and treatment of mental health problems in children.

    - Specifically, Healthy People 2010 states as goals:

        18-2.1. Reduce the rate of suicide attempts by adolescents (Solution: "Reduction in access to lethal methods and recognition and treatment of mental and substance abuse disorders are among the most promising approaches to suicide prevention.")

18-7.1. Increase the proportion of children with mental health problems who receive treatment. ("Better services and collaboration for children with serious emotional disturbance and their families will result in greater school retention, decreased contact with the juvenile justice system, increased stability of living arrangements, and improved educational, emotional and behavioral development [63,64]."

18-8. Increase the proportion of juvenile justice facilities that screen new admissions for mental health problems.

## 1. Statement of the Research Question

Does early identification and treatment of maternal depression influence infant and toddler outcomes?

## 2. Hypothesis

MH-1a) Early identification and evidence-based treatment of maternal depression will be related to better infant and toddler functioning (age-appropriate learning and problem solving, communication, mobility, self-care and socialization).

MH-1b) Communities with enriched early family intervention programs, those with high levels of social capital and those with supplemented child care programs will have better outcomes for infants and toddlers with depressed mothers.

## 3. Rationale and Significance

Approximately 10-15% of women experience depression during pregnancy and/or within the first six months following childbirth (Lumley & Austin, 2001, Ferguson et al., 1996). When untreated, perinatal and postpartum clinical depression can result in serious dysfunction and disability. Apart from the adverse consequences for women themselves, there is the possible negative impact of maternal depression on the relationship between the mother and her family, and on her child's emotional, behavioral and cognitive development (Civic et al. 2000).

Most women experiencing depression do not seek professional help despite the fact that they often have contacts with health professionals (e.g., internists, obstetricians, pediatricians) during pregnancy and following birth. Almost 50% of those in need of treatment do not seek help from family or friends (Small et al. 1994). Recognition of maternal depression in primary care is poor (Barnett et al. 1993), which has led to increasing interest in the use of screening, either antenatally or postnatally.

For women who do seek help, efficacious treatment is available. Several studies have demonstrated the efficacy of antidepressant medication for moderate to severe depression, cognitive-behavioral and interpersonal psychotherapy for less severe depression, and a combination of medication and crisis intervention and support for suicidal ideation and psychosis (Miller, 2002).

## 4. Implications of Hypotheses

### a) Study Design

Regular, fairly frequent assessments- every 3 to 4 months Year 1, 2 per year up to age 3. Need to cluster the sample and insure service system variation among sites. Without clustering, service systems would be too numerous to measure.

**b) Sampling Strategy**

Ideally, we need to augment interviews of high-risk pregnant women (those who are poor, single, without social support, with prior mental health problems) and a sample of those without risk factors to measure maternal depression, services and child outcomes.

**c) Measurement**

Need to measure maternal depression, service use and infant/ toddler outcomes. Would need chart reviews or pharmacy records to determine SSRI dose, duration. Would need to gather contextual data from public use sites and governmental publications to assess community resources.

## 1. Statement of the Research Question

Can we identify early indicators of mental health problems within the medical, childcare and educational systems?

## 2. Hypothesis

MH-2a) Early identification in the disease process for identified mental health issues (those with efficacious treatments) is more likely to lead to treatment.

MH-2b) Early identification will vary by family, child, practitioner, health care system and community characteristics, including post trauma responses to bioterrorism and other disasters.

MH-2c) Early assessment of parental attitudes, ability to act as child advocates and willingness to seek help would identify the most important early opportunities to make families better prepared to seek and obtain treatment.

## 3. Rationale and Significance

In chronic conditions of childhood, parental preferences, attitudes and beliefs may be the most important indicators of parental identification, help seeking and adherence with treatment.

## 4. Implications of Hypotheses

### a) Study Design

Need a clustered sample so that provider and service system characteristics can be measured. Ideally, sites would have some community/ service system variation.

### b) Sampling Strategy

Need to augment interviews of high-risk families (poor, single parents, prior mental health problems) to gather detailed service use and parental attitudes.

### c) Measurement

Need measures for child mental health issues that work across the life span; need measures of service system characteristics as well as barriers and stigma.

## 1. Statement of the Research Question

Does evidence-based care for key identified mental health problems improve child outcomes?

## 2. Hypothesis

MH-3a) Evidence-based care for key identified mental problems delivered within routine clinical practice will improve child functioning.

MH-3b) Receipt of evidence-based care will vary by out-of-pocket costs, parental attitudes and community characteristics.

## 3. Rationale and Significance

Within the last eight years, evidence-based care has been developed for selected emotional and behavioral problems. Use of such care and effectiveness of such care is unknown within routine community service settings.

## 4. Implications of Hypotheses

### a) Study Design

Need to cluster sample since practice records will need to be searched and communities will need to be described.

### b) Sampling Strategy

Same as Hypotheses 1 and 2.

### c) Measurement

Need to identify/ develop measures of evidence-based care for selected conditions.

## 1. Statement of the Research Question

Do organizational and financing features affect access to and qualify of mental health treatment?

## 2. Hypothesis

MH-4) Organization and financing features of children's primary health care and mental health services are related to access, quality and cost.

## 3. Rationale and Significance

Attempts to investigate organization and financing issues have been unsuccessful due to insufficient data. The NCS has the potential to provide data to answer these critical questions.

- The children's mental health system is complex and fragmented, with separate mental health services having different pathways of entry and funding streams.
- As highlighted in the recent IOM report "Crossing the Quality Chasm," the structure of the health care systems and processes within them make attainment of high-quality care difficult.
- This study, with the tracking of 100,000 pregnancies, has the potential to examine a wide variety of organizational and financing features of the health care systems. May be able to determine to what extent and in which areas the organization, financing and provision of health care acts as a negative (harmful) or positive (beneficial) exposure/mediator/moderator on child functioning.

## 4. Implications of Hypotheses

### a) Study Design

Same as previous hypotheses

### b) Sampling Strategy

Same as previous hypotheses

### c) Measurement

Need measures of the content of insurance policies, public payment regulations and organizational features for key services.

## Preventive Services in the National Children's Study

### Introduction

The subcommittee has considered detailed hypotheses related to preventive services that span the age spectrum from early childhood (with heavy emphasis on behavior and development) to adolescence. However, given that the National Children's Study will likely focus on key conditions and certain overarching hypotheses, the subcommittee chose to develop two broad research questions that could be molded to fit with the as of yet undetermined, broad study guidelines.

In addition, the Preventive Services group identified that a strength of the NCS would be to study the effectiveness of implementing in a large population sample those preventive services for which there is the strongest evidence base. In addition, an import component of the NCS could include the study of newer screening tests that have been proven to be efficacious in pediatric clinical settings (e.g., screening for maternal depression); this study would allow the testing of the effectiveness of these interventions in large populations of children, since the impact of effectiveness due to variations availability of community resources and provider awareness of such resources is not known.

The group also believed that nested studies could be used to: 1) test models of care delivery for preventive services; 2) examine critical and sensitive periods for providing preventive and promotional services; 3) identify predisposing attributes that relate to the adoption of healthy behaviors among parents and their children.

A major, cross cutting health services question, though not specific to preventive services, is the examination of whether current measures of quality of care are associated with improved health outcomes for children. This appears more of an issue for interpersonal, rather than technical, measures of quality. For example, improvements in interpersonal quality may lead to adherence to recommendations, but may not lead to sustained behavior change, and ultimately, improved health of children. We did not include this as a central preventive services question.

As second, large cross cutting health services question, again not specific to preventive services, is understanding how the organization and financing of child health care services contributes to changes in services delivery/accessibility, unmet needs of children, quality of care, and adoption of health promoting and risk behaviors

Finally, we agreed that the NCS provides a unique opportunity to provide descriptive information regarding:

--trajectories of parenting practices over time as mediators of health services leading to optimal health behaviors/practices by children.

--trajectories of externalizing behaviors and development of other psychopathology

--determinants of social health of children through assessments of parenting practices, community resources, educational exposures, social environment, and genetic/biological predisposition

--describing natural history of evolution of population of CSHCN

**Research Question 1:** Does universal screening improve health outcomes for children?

**Hypotheses**

PS-1a) Expanded, mandatory newborn screening (e.g,. screening for CF and other conditions soon to be identified through human genome project) does not improve health of children.

PS-1b) Behavioral and developmental testing is optimized when critical/sensitive time periods are identified.

PS-1c) Selected screenings for targeted populations are highly effective.

PS-1d) Universal screenings for selected conditions are highly effective.

**Rationale**

A variety of universal screens are recommended for children throughout childhood despite modest evidence base of the effectiveness. These screenings include, but are not limited to: mandatory newborn testing (e.g., galactosemia, sickle cell, cystic fibrosis in some states); U/A; anemia; vision/hearing; behavior/development; psychosocial assessment; domestic violence; testicular self examination; blood pressure; and maternal depression. Considerable time, cost and resources are allocated and recommended, universal screens are not consistently provided. Variation in screening may contribute to health disparities and poor quality of care.

The large cohort study would provide unique opportunity to consider screenings, in particular, for low frequency outcomes with severe consequences as well as outcomes that have higher frequency and modest individual impact though may have significant cumulative impact on a population of children.

The large cohort also would provide a unique opportunity to examine the impact of state policies regarding newborn and other screenings on child health (e.g., does screening for select conditions in newborn period, beyond core set, actually improve health outcomes for those children with early identification?). Additional rationales were provided for more specific hypotheses included in the meeting binders.

**Implications for Study Design**

Specific screening tests should relate to marquis hypotheses and sentinel conditions, to the extent feasible.

Determining whether screening is provided could require chart review (e.g., anemia screening), review of state newborn screening records linked to vital statistics data (e.g., mandatory newborn screening), or possibly parent recall shortly following a visit.

Selected outcomes would vary by screening test. Thus, for behavior and development, outcomes might include measures related to family responsiveness, school preparedness, referrals to community services (e.g., early intervention or educational programs, parenting services, mental health specialists), diagnosis and treatment of behavioral problems and disorders, social skills.

There is considerable interest among this subgroup to examine community level variables such as infrastructure for health care delivery and related services.

Peter Margolis suggested a pilot study related to decision modeling to examine issues related to test, disease, and treatment characteristics associated with improved health outcomes. (He planned to send a separate paragraph describing this proposed pilot).

**Research Question 2:** Does anticipatory guidance improve health outcomes for children?

Hypotheses:

PS-2a) Office based anticipatory guidance appears most effective when broad based community approaches are used to improve health related behaviors.

PS-2b) Office based anticipatory guidance has varying effectiveness for subsets of the population at critical/sensitive time periods.

Rationale:

As evidenced by Bright Futures, a broad array of topics has been identified as important to consider during health supervision. These recommendations, although intended to be individualized to meet the needs of specific children and families, are largely universal. Many recommendations are not implemented due to limited time, staffing shortages, resource constraints, provider doubtfulness about efficacy, managed care constraints, and insufficient reimbursement concerns.

Beyond selected, proven efficacious counseling for specific injuries and other selected preventive health behaviors, we know relatively little about how to increase the effectiveness of anticipatory guidance. Yet, preventable conditions, specifically injuries, are the leading cause of mortality of children in the US, and risk behaviors are a major cause of morbidity (e.g.., obesity, smoking, STDs). Our inability to change behavior has contributed enormously to health care costs as well as the burden of disease for selected conditions.

Child health providers share in the credit for efforts to change parenting behavior with regard to sleep position; yet, it would appear that messages provided by child health providers are most successful in the setting of community wide social marketing and public health campaigns, such as the Back to Sleep initiative. Moreover, despite successful passage of state and county laws to regulate risk-taking behaviors (e.g., failure to use seat belts, bike helmets, car seats), there remains room for substantial improvement (e.g., increase % correctly installed car seats, increase in % infants using correct sleep position).

Implications for Study Design:

As for hypothesis 1, specific anticipatory guidance topics should relate to marquis hypotheses and sentinel conditions, to the extent feasible.

Outcomes could include parenting practices (e.g., discipline techniques, use of routines, foods served at meal time or in refrigerator/cupboard), child health outcomes (e.g., injuries, exposure to reading, body mass index, developmentally appropriate physical activity), and health care utilization (e.g., use of ED for behavioral/developmental concerns).

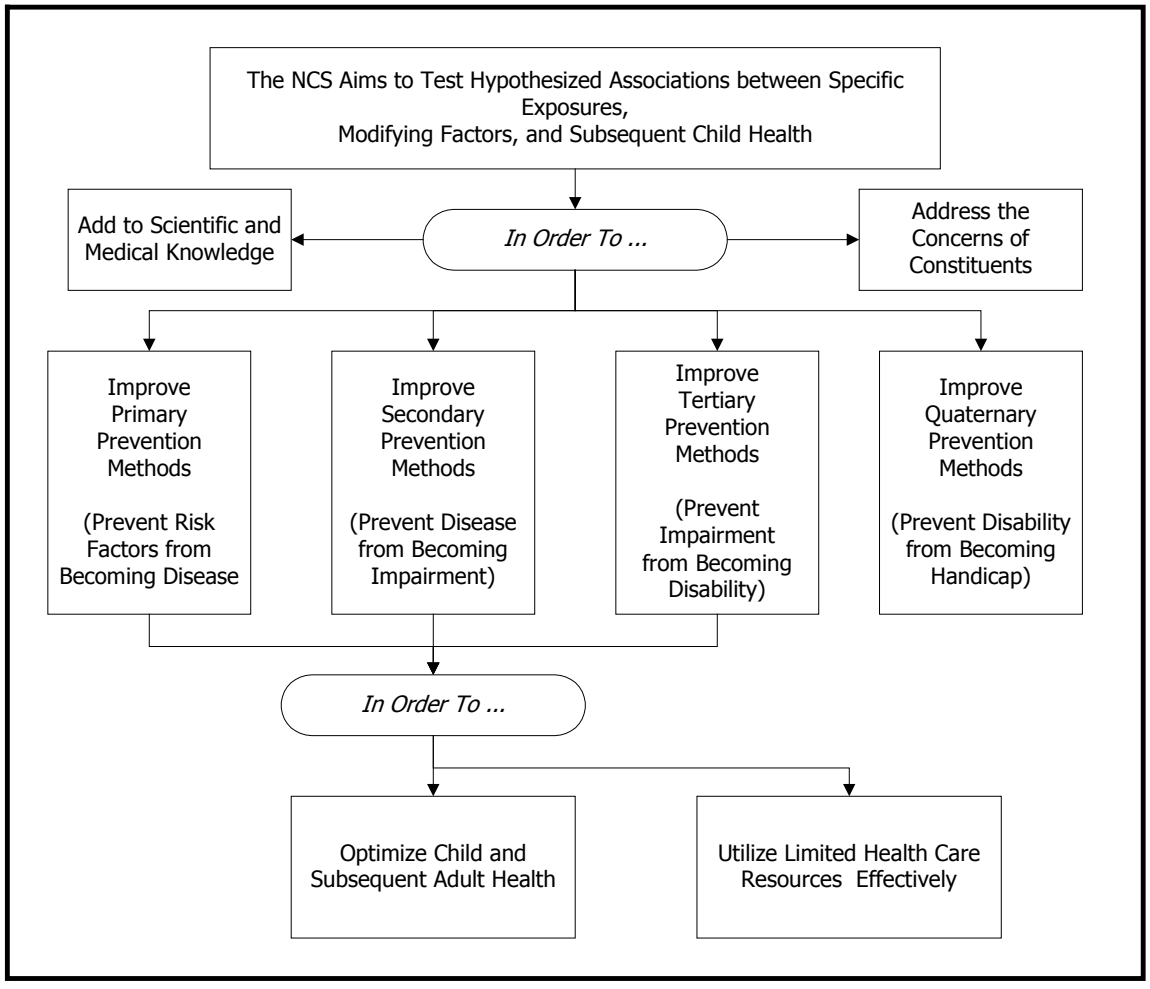
Corresponding data collection tools could include parent interviews, self report for older children/adolescents, medical chart reviews, physical assessments, and review of administrative records.

A pilot study regarding understanding the validity of parental report regarding many of these measures is recommended to address issues regarding the duration of the accuracy of recall (?12 months for health care utilization as in existing, national surveys), recall regarding the type and content of pediatric visits for the specified anticipatory guidance areas to be examined, and understanding whether parental report correlates with improved child health outcomes.

## Medical Care Services in the National Children's Study

### Introduction

We understand the aim of the NCS is to improve the health of children (and subsequently, the adults that children become) by furthering our understanding of how specific exposures impact child health, and how the impact of these exposures are modified by either risk or protective factors. In order for this basic knowledge of exposures and of modifying risk or protective factors to be translated into improved child health outcomes, the knowledge will need to guide improvements in our methods of health promotion and of primary, secondary, tertiary, and quaternary prevention (designed, respectively, to target the progression from risk factors to impairment to disability to handicap – See Figure 2). If the study also illuminated how to makes these improvements in a cost-effective manner, all the better. The diagram below makes explicit the goals of the study:




Health services can play a key role modifying the interactions between environmental exposures (infectious and non infectious) and host/genetic determinants that impact child health outcomes.

Therefore, including a robust health services research component in the NCS will be critical to the success of this project.

## IMPACT OF HEALTH SYSTEM CHARACTERICISTICS ON IMMUNIZATION DISPARITIES

### Background

Immunization is one of the most effective and cost-effective means to promote the health of children, yet many pockets of unimmunized children especially among low-income urban children remain throughout the country. In addition concern is being raised about the safety of routine vaccination and possible neurodevelopmental sequelae. It is important to address the science and perception of safety issues especially with regard to thimerosal and MMR as well as provider and parent perception of risks and benefits of vaccines including new vaccines. We also need to assess methods of reducing the pockets of under-immunization including the effectiveness of electronic registries, public health interventions, and state and federal child health vaccine policies.

### Hypothesis

MC-1) The proportion of children who have up-to-date immunization status will be greater in health care systems that utilize electronic medical records (EMR). Sub-hypotheses include:

- Effect of EMR will be greater for newly recommended compared to established immunizations.
- Effect of EMR will be greater for historically-under-immunized groups of children.

### Rationale and Significance of Hypothesis

With a host of new immunizations on the horizon, and known difficulties in the universal delivery of the current immunizations, the NCS could add substantially to understanding of why certain children do not receive certain immunizations, and potentially identify systems that effectively overcome barriers.

### Hypothesis Testing Implications Regarding Study Design and Methods

If data is collected a sufficient degree of detail, features of different EMR systems could also be examined.

A2-stage sampling strategy could be used to efficiently gather the necessary information, noting in the first stage simply whether a EMR is used in the child's health care system, and in the second stage collecting more detailed information about the EMR system.

IMPACT OF GENETIC MODIFICATION OF RISK OF VACCINE-RELATED ADVERSE EVENTS

**Background**

Vaccine-related adverse events are of concern to children, parents, providers, and others who wish to have the safest and most-effective immunization policy possible. Little is known about whether, and how, individual risk of vaccine-related adverse events varies.

**Hypothesis**

MC-2) Genotypic differences across individuals modify the risk of post-immunization known and hypothesized adverse events, including seizures and the development of autism.

**Rationale and Significance of Hypothesis**

The NCS is ideally suited, because of its protection against selection bias due to its prospective design, with possible banking of blood samples for multiple purposes, to address this question.

Given the importance of immunization to the health of children and our nation, and the need to minimize immunization-associated risk in order to maximize the benefits of individual immunization and of immunization policy across the public, the identification of specific genetically-at-risk profiles would enable basic and health services research to devise strategies to minimize this risk.

**Hypothesis Testing Implications Regarding Study Design and Methods**

The number of febrile seizures attributable to the administration of DTP and MMR vaccines was estimated to be 6 to 9 and 25 to 34 per 100,000 children, respectively (Barlow and others, 2001).

1 in 1,000 individuals diagnosed the "classic" autism; 1 in 500 individuals within the Autism Spectrum, including Pervasive Developmental Disorders (PDD); and 1 in 200 individuals within the Autism Spectrum, including PDD and Asperger's Syndrome.

Nested case-control methods could be used within the larger cohort design to retrieve banked samples of genetic material from cases (identified prospectively as have immunization-related seizures or other adverse events) and controls, with either gene-array exploratory analysis or hypothesis testing of candidate risk-modifying genes.

IMPACT OF GENOMIC INFORMATION ON CHILDREN & FAMILIES

**Background**

In the years to come, many children will be identified as having genotypes that put them at risk for undesired health conditions or that place them in a physiologically-distinct subgroup of a particular disease state. With this information, children will likely be recommended to receive ongoing medical surveillance, or preventative treatments to reduce their risk of subsequent disease, or specific modification of disease management in keeping with their genetically influenced physiology. The beneficial and adverse consequences of such genomic information and subsequent health care response to this information remain unknown.

**Hypothesis**

MC-3) Learning of a child's genomic information regarding risk and specific disease states will benefit the child to the degree that they are cared for by health care providers who are expert in the use of genomic information.

ALTERNATIVELY …

Knowing 'risk' will increase child and parental anxiety, health care utilization, and cost without discernible benefit. This relationship will be modified by:

- The child's age
- The patient-centered-ness of medical care
- The immediacy of perceived risk
- The efficacy of available interventions against the target disorder

[XXX Alternatively #2 ala WFL:

The degree of benefit of risk information obtained from genomic information for a child will be a dependent on the availability of preventive and/or therapeutic options to manage genetic risk and the degree to which the child's providers have expertise in the use of genomic information. This information may overall be harmful to the extent that the information increases child and parental anxiety, health care utilization, and health care costs in the absence of well-defined and well-communicated risk management options. The overall benefit of this genomic information will be modified by:]

**Rationale and Significance of Hypothesis**

Taking the genomic revolution from the bench to the bedside, with tangible benefits for patients, will require that individual level genomic information be used with an understanding of its implications and limitations, within a health care system that can mount the appropriate interventions.

ALTERNATIVELY …

This early detection will become increasingly common, will cost a great deal, and will be associated with variable adherence to recommended interventions and therefore may not have substantial benefit.

**Hypothesis Testing Implications Regarding Study Design and Methods**

From the larger cohort of children, we will need to be able to identify cases for whom genomic risk status or disease grouping has been created. These cases could then either be followed prospectively, or assessed retrospectively. Will need to measure health status, including mental health, utilization, and costs.

IMPACT OF BEREAVEMENT CARE ON SIBLINGS AND PARENTS OF CHILDREN WHO DIE

**Background**

Although expert opinion and case series from individual institutions suggest that bereavement care is beneficial to siblings and parents who have experienced the death of a child in their families, little is know about the extent of these services or their impact on family members. Exposure to childhood death, while infrequent, is not rare. In the United States in the year 2000, a total of 53,728 children and adolescents between birth and 19 years of age died. The infant mortality rate was 6.9 deaths per 1000 live births. For 1998, the fetal death rate was 6.7 fetal deaths per 1000 live births and fetal deaths. For 2000, the annual rate of death for children age 1-19 years was 34.5 deaths per 100,000 persons per year (Hoyert and others, 2001).

**Hypothesis**

MC-4) Following the death of a child, siblings and parents who receive bereavement care have better outcomes than those who do not. Sub-hypotheses include:

- Impact of bereavement care for siblings will vary by the sibling's developmental age.
- Impact of bereavement care for parents will vary by the age of the deceased child and the manner of that child's death (acutely traumatic versus terminal chronic illness).

**Rationale and Significance of Hypothesis**

Several studies have documented the negative health impact of having a sibling or child die.

Although methods of bereavement care could be studied in the framework of RCTs, such study is unlikely to be used for the vast majority of services being offered. If the NCS could show a benefit, then improvements in services could be subjected to more rigorous study.

**Hypothesis Testing Implications Regarding Study Design and Methods**

In a cohort of 100,000 pregnancies, if current rates continue, we should expect approximately 670 fetal deaths and 688 infant deaths, with approximately 34 deaths per year thereafter (with more during early childhood and late adolescence, and less in between).

The index subject need not be the one who died; the subject may be the sibling of another child who died in the family.

We do not know the proportion of family members of these fatalities that receive bereavement services. A pilot study could address this unknown parameter.

Health status, including mental health, and utilization and cost information would need to be collected for the sibling and the parents.

IMPACT OF PROVIDER AND HOSPITAL VOLUME ON PEDIATRIC SURGICAL OUTCOMES

**Background**

In the realm of adult medicine, evidence is mounting that surgical outcomes are superior when the surgeon and the hospital have greater experience in performing the target procedure.

**Hypothesis**

MC-5) Surgical outcomes are superior for children who are cared for in children's hospitals than children cared for in community hospitals.

**Rationale and Significance of Hypothesis**

These findings would inform debates over the regionalization of tertiary pediatric health services, with aggregation of cases in regional centers in order to increase patient volumes and ultimately to improve outcomes.

**Hypothesis Testing Implications Regarding Study Design and Methods**

Ideally, would have pre-operative health status details in order to adjust for severity. Would also want to have as much information regarding utilization (such as LOS) and subsequent outcome (health status) as possible.

IMPACT OF A MEDICAL HOME FOR CHILDREN WITH SPECIAL HEALTH CARE NEEDS

**Background**

Many national experts in pediatric health care, including the American Academy of Pediatrics, believe that the medical care of infants, children, and adolescents ideally should be accessible, continuous, comprehensive, family-centered, coordinated[1], compassionate, and culturally effective[2]. It should be delivered or directed by well-trained physicians who provide primary care[3], and manage and facilitate essentially all aspects of pediatric care. The physician should be known to the child and family and should be able to develop a partnership of mutual responsibility and trust with them. These characteristics define the "medical home" and describe the care that has traditionally been the goal of community physicians in the office setting. In contrast, care provided through emergency departments, walk-in clinics, and other urgent-care facilities is often less effective and more costly[4].

The US Department of Health and Human Services "Healthy People 2010" goals and objectives state that "all children with special health care needs will receive regular ongoing comprehensive care within a medical home."[6] The Federal government has recognized the importance of a medical home by implementing the Title V Maternal and Child Health Program and requiring each state to report annually on the number of children with special health care needs who have access to a Medical Home.

Proponents of the medical home concept for children with special health care needs (CSHCN) maintain that children who have a medical home receive better, more comprehensive care, that is cost-effective, and improves outcomes for CSHCN, their siblings, and parents.

**Hypothesis**

MC-6) CSHCN who have a medical home are more likely to receive comprehensive preventive and acute health care. CSHCN with a medical home have improved health outcomes and the care provided is more cost effective. Sub-hypotheses to be tested include:

- Health care systems that employ electronic medical records will better coordinate care for CSHCN.
- Children in health care systems with defined relationships with schools and other community resources will be more likely to have a medical home and improved outcomes.

**Rationale and Significance of Hypothesis**

- Although the potential benefits of enhanced care coordination could be tested through a RCT, most systems would be unwilling to randomize children to the 'usual care' arm.
- Conceivable that investments in care coordination would be cost-savings.
- The study will also collect important descriptive information regarding the eight components of a medical home: 1) Accessible 2) Family-Centered Compassionate 3)

Continuous 4) Comprehensive 5) Care Coordination 6) Culturally Effective 7) Have a primary care provider 8) Have a usual source of care. This information then will be used to examine specific, predictive models for quality care and improved outcomes.

**Hypothesis Testing Implications Regarding Study Design and Methods**

We feel that the most reliable and valid source of data for whether a child has a special health care need and for whether a child has a medical home should come primarily from patient-based measures including surveys/interviews to/with the parents of children unable to respond for themselves and then eventually teen-based measures.

In order to obtain sufficient sample sizes, we recommended that a non-categorical definition of children with special health care needs be used that is consequences based rather than condition-specific.

Requires that there be sufficient variation in the degree of care coordination across the sub-cohort of CSHCN in order to control for potential confounders yet still discern an impact of care coordination.

If during the duration of the NCS, individual CSHCN experience a change from a coordinated system to an uncoordinated system, or vise-versa, then an interrupted time-series design would be feasible.

Otherwise, observing differences across CSHCN, with adjustment for child characteristics, would be the essential design.

IMPACT OF A MEDICAL HOME ON CARE AND HEALTH OUTCOMES FOR ALL CHILDREN

**Background**

Multiple federal programs require access to an ongoing source of health care. Additionally, the "Future of Pediatric Education II" goals and objectives state, "Pediatric medical education at all levels must be based on the health needs of children in the context of the family and community" and that "all children should receive primary care services though a consistent 'medical home'."[7] Over the next decade, with the collaboration of families, consumers, government, medical education, and the health care industry, we can improve the quality of life for all children in the community through the care provided in a medical home.

The American Academy of Pediatrics defines a medical home for infants, children, and adolescents as a care system that is accessible, continuous, comprehensive, family-centered, coordinated[1], compassionate, and culturally effective[2]. Furthermore, it defines recommends that care should be delivered or directed by well-trained physicians who provide primary care[3], and manage and facilitate essentially all aspects of pediatric care. The physician should be known to the child and family and should be able to develop a partnership of mutual responsibility and trust with them.

Medical care is at times provided in locations other than physicians' offices, such as hospital outpatient clinics, school-based and school-linked clinics, community health centers, health department clinics, and others. Regardless of the venue in which the medical care is provided, to meet the definition of "medical home" a physician must ensure that the services listed above are provided.

**Hypothesis**

MC-7) Children who have a Medical Home are more likely to receive comprehensive, quality health care that achieves cost-effective health care and improved health outcomes for the child and family. A sub-hypothesis:

- Children in health care systems with defined relationships with schools and other community resources will be more likely to have a medical home and improved outcomes.

**Rationale and Significance of Hypothesis**

Children who have a Medical Home will be more likely to receive comprehensive quality of care which will in turn lead to improved health outcomes and cost effective care. We have seen in past research that children who have a personal doctor or nurse, one component of the medical home, are more likely to receive specific aspects of recommended care and parents are more likely to report doing certain activities with their child to help them grow and learn.

These findings will be significant because they will note and evaluate specific aspects of the health care system (eight components and one composite measure of Medical home)

that will be predictive of quality care and improved health outcomes. Policy implications for how systems and care can be improved will be noted.

**Hypothesis Testing Implications Regarding Study Design and Methods**

Data regarding whether a child has has a medical home and about certain aspects of health care received should come primarily from patient-based measures including surveys and/or interviews to/with the parents of children unable to respond for themselves and then eventually teen-based measures. Included in these data collection efforts should be an effort to measure health outcomes of the parent and the efficacy with which they are able to care for their child and maneuver through the health care system.

IMPACT OF CHILD CARE CENTER AND SCHOOL BASED PROGRAMS ON OBSEITY RISK REDUCTION

**Background**

Obesity among children has reached epidemic proportions. Prevalence varies markedly across socioeconomic strata. A recent Cochrane review of programs to prevent obesity noted that most studies have shown no benefit, with the few studies showing effectiveness focusing on promotion of physical activity.

**Hypothesis**

MC-8) Children who attend child care centers and schools that have comprehensive, enjoyable programs that promote physical activity and active lifestyles will have lower BMIs and a higher proportion of children that exercise when compared to children attending centers or schools without such programs.

**Rationale and Significance of Hypothesis**

Schools are the only place for population-based intervention opportunities for enjoyable physical activity everyday.

These findings will be significant as there are few interventions that have been proven to be effective in reducing infant, child, or adolescent obesity.

**Hypothesis Testing Implications Regarding Study Design and Methods**

We feel that the most reliable and valid source of data related to obesity is from surveys to or interviews with the parents of children unable to respond for themselves and then eventually teen-based measures.

Exercise should be defined as whether the child or teen engages in physical activity for 30 minutes, 5 days a week in school.

Enjoyable physical activities will be defined and measured by the child or teen.

## Dental Services in the National Children's Study

### Introduction

Despite overall improvements in oral health during the past several decades, tooth decay remains the most common chronic disease of childhood, affecting 5-8 times as many children as asthma.[1] By mid-childhood over 54% of children have detectable dental caries, and by late adolescence about 80% have acquired this preventable infectious disease.[2, 3]

Glaring disparities in children's oral health and access to dental services have been reported by the U.S. General Accounting Office (GAO), U.S. Surgeon General and others.[4, 5, 6] Low-income and minority children and those with special health care needs (CSHCN) are at greatest risk of inadequate access and poor oral health.[4, 5, 6, 7] The magnitude of the problem is such that analyses of national data have identified dental care as the most prevalent unmet health need in U.S. children.[8]

Childhood oral diseases have significant consequences for health, development, self-esteem, social function and well-being that, until lately, have received little attention. Recent scientific studies also have highlighted relationships among mothers' oral health and pregnancy outcomes as well as the oral health of their children.[6]

Accordingly, children's oral health has been recognized as an essential element of children's general health and an important area within children's health services research. The research questions and hypotheses outlined below are seen as priority issues for investigation within the context of the National Children's Study.

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3 National Institute of Dental Research. *Oral Health of the United States Children: The Nationals Survey of Dental Caries in School Children, 1986-87; National and Regional Findings*. Bethesda, MD: National Institute of Dental Research, 1989: DHHS publication no. (PHS) 89-2247.

4 Vargas CM, Crall JJ, Schneider DA. Sociodemographic distribution of pediatric dental caries: NHANES III, 1988-1994. *J Am Dent Assoc*. 1998;129:1229-1238.

5 U.S. General Accounting Office. *Oral Health: Dental Disease is a Chronic Problem among Low-income Populations.* April 2000. GAO/HEHS-00-72 www.gao.gov (accessed July 31, 2002)

6 U.S. Department of Health and Human Services. Oral Health in America: A Report of the Surgeon General. Rockville, MD: U.S. Department of Health and Human Services, National Institute of Dental and Craniofacial Research, National Institutes of Health, 2000.

7 Newacheck P, McManus M, Fox HB, Hung Y, Halfon N. Access to health care for children with special health care needs. *Pediatrics* 2000; 105: 760-766

8 Newacheck PW, Hughes DC, Hung YY, Wong S, Stoddard JJ. The unmet health needs of America's children. *Pediatrics*. 2000;105(4 pt 2):989-997.

1. **Statement of the Research Question**
   - **a.** Do children who receive recommended well-child services provided by primary medical care providers have lower risk of developing dental caries compared to children who do not receive recommended well-child services?
   - **b.** Do children who receive recommended well-child services from primary medical care providers have higher rates of dental service utilization compared to children who do not receive recommended well-child services?

2. **Hypothesis**

   DH-1a) Children's risk of developing dental caries is inversely related to the degree to which they receive recommended well-child services from primary medical care providers.

   DH-1b) Children's utilization of dental services is positively related to the degree to which they receive recommended well-child services from primary medical care providers.

3. **Rationale and Significance of the Hypothesis and Relevance to the Research Question**

   - **a.** Children under 3 generally have access to primary medical care providers, but not dental providers.
   - **b.** Professional guidelines (e.g., U.S. Preventive Services Task Force, Bright Futures, Medicaid/EPSDT Guidelines) recommend that primary care clinicians provide oral health assessments and counseling and refer children in need of treatment to dental care providers.

4. **Implications of Hypotheses**
   - **a. Study Design:** The longitudinal nature of this study is ideally suited for this research question.
   - **b. Sampling Strategy:** The sample needs to include children from different socioeconomic strata.

   - **c. Measurement**
     - i. Putative risk factors need to be assessed periodically.
     - ii. Knowing whether specific attention was directed to dental or oral health during well-child visits would strengthen the analysis of the hypothesized relationship.

   - **d. Other Implications for Consideration:** Risk for developing dental caries is multi-factorial and related to social, community and family characteristics that may confound the analysis and for which data need to be obtained and controlled.

### 1. Statement of the Research Question

**b.** Do children who receive early and regular oral health services from dental care providers have lower risk of developing dental caries and lower incidence of dental caries compared to children who do not receive early and regular dental services?

**c.** Do children who receive early and regular oral health services from dental care providers have less untreated dental disease, lower overall dental and health care costs, and improved oral health-related quality of life compared to children who do not receive early and regular dental services?

## 2. Hypothesis

DH-2a) Children's risk of developing dental caries is inversely related to the degree to which they receive early and periodic oral health services from dental care providers.

DH-2b) Children who receive early and regular oral health services from dental care providers have less untreated dental disease, lower overall dental and health care costs, and improved oral health-related quality of life compared to children who do not receive early and regular dental services.

## 3. Rationale and Significance of the Hypothesis and Relevance to the Research Question

**a.** Dental caries remains the most common chronic disease of childhood and occurs with relatively high prevalence in infants and very young children.

**b.** The incidence and severity of childhood dental caries can be reduced through a combination of professional services and self/parental care activities.

## 4. Implications of Hypotheses

**a. Study Design:** The longitudinal nature of this study is ideally suited for this research question. Little information currently exists on the differential impact of common preventive services for children having different levels of risk.

**b. Sampling Strategy:** The sample needs to include children from different socioeconomic strata.

**c. Measurement**

**i.** Putative risk factors and oral health status need to be assessed periodically, ideally by a clinical examination

ii. Information on the timing and the content of dental services will be required, ideally from administrative data or clinical records.

**d. Other Implications for Consideration:** Risk for developing dental caries is multi-factorial and related to social, community and family characteristics that may confound the analysis and for which data need to be obtained and controlled.

## 1. Statement of the Research Question

**a.** Do children with medical and/or dental insurance coverage have increased access to and use of oral health services compared to children who lack medical and/or dental insurance coverage?
**b.** Does the quality of children's medical and/or dental insurance affect their access and use of dental services?
**c.** Do children whose parents have medical and/or dental insurance coverage have increased access to and use of oral health services compared to children whose parents lack medical and/or dental insurance coverage?

## 2. Hypothesis

DH-3a) Children covered by medical and/or dental insurance are more likely to receive dental services.

DH-3b) The quality of children's medical and/or dental insurance is related to the extent to which they receive dental services.

DH-3c) Children whose parents have medical and/or dental insurance coverage have increased access to and use of oral health services compared to children whose parents lack medical and/or dental insurance coverage.

## 3. Rationale and Significance of the Hypothesis and Relevance to the Research Question

**a.** Prior studies have shown that reducing cost barriers leads to higher rates of utilization.
**b.** For every child who lacks medical insurance, there are 2.6 children who lack dental insurance.
**c.** Problems of access to dental services, especially for low-income children, have highlighted the importance of quality dental insurance coverage (e.g., through SCHIP and Medicaid programs).

## 4. Implications of Hypotheses

**a. Study Design:** The longitudinal nature of this study is ideally suited for this research question.

**b. Sampling Strategy:** The sample needs to include children from different socioeconomic strata.

**c. Measurement:** Being able to characterize the nature of the child's dental and medical insurance coverage is critical.

## 1. Statement of the Research Question

**a.** Do children with cleft lip and/or cleft palate who are treated by craniofacial teams have better health outcomes than children not treated by craniofacial teams?

## 2. Hypothesis

DH-4a) The timing and quality of health services for cleft lip and/or cleft palate affects outcomes.

DH-4b) Children with cleft lip and/or cleft palate who are treated by craniofacial teams have better health outcomes than children not treated by craniofacial teams.

## 3. Rationale and Significance of the Hypothesis and Relevance to the Research Question

**a.** Cleft lip and/or cleft palate remains a relatively common birth defect (1:600 – 1:900) that has substantial quality of life implications and requires an extensive array of health care services.

## 4. Implications of Hypotheses

**a. Study Design:** The longitudinal nature of this study is ideally suited for this research question.

**b. Sampling Strategy:** None.

**c. Measurement:** None.

## Community Health Services in the National Children's Study

### Introduction

1] The World Health Organization definition of health is utilized in formulating hypotheses around the role of community health services.

2] The significance of community health services lies in two basic realities:

a) The importance of social and community determinants of health and well-being for children: the significance of *social context*.
b) The limitations of our health system which focuses primarily on individuals and not populations.

3] In spite of the use of a broad definition of health, and recognition of the importance of social as well as biologic determinants of health, there still are uncertainties in what is a "health outcome". For example, many would suggest that high school dropout is primarily an educational outcome, or that incarceration of teenagers is an issue of juvenile justice. In our view, however, both of these issues have immense implications for the health and well-being of children and youth, and are legitimate "health outcomes".

4] The definition of "community health services" used by us is as follows: Services that fall out of traditional individual health care provider or traditional institutional health care provider settings that affect the health and well-being of children (hopefully but not necessarily in a positive direction).

5] To test hypotheses concerning community health services, it would be preferable to measure community-level variables directly, e.g., actually assessing the characteristics and availability of community Head Start programs and public health media campaigns. For the purposes of research design for The National Children's Study, it may be necessary to measure "what is used" or "what is perceived to be available" at the individual-level instead.

6] A major problem exists in the determination of efficacy of given community health services, particularly the efficacy of those services that vary significantly based on local and regional characteristics. Expressed differently, it has not been possible to subject many if not most community health services to the kind of controlled studies that are required to clearly demonstrate efficacy. Similarly, with few exception controlled studies in the community have not been replicated in communities with different characteristics.

7] If a significant component in the study of the effect of community health services requires careful assessment of community structure and function, a cluster analysis methodology will need to be employed.

8] Since many communities will introduce or eliminate community services programs during the course of the study, consideration might be given to intake a cohort in year 1 and another cohort in year 3 or 4 rather than intake continuously. Although it would increase the cost of the study, it

also would increase the opportunity to determine impact of change in services on health outcomes.

9] Our strategy is to ask what is the role of defined community health services on specific outcomes or markers. These outcomes, as developed below, have varying clarity of identity as a "health" outcome.

Four health outcomes have been selected: asthma, obesity, retention in 1st grade, and initiation of tobacco smoking. A series of community health services or activities is listed below for each of these health outcomes. In the attached sheets, the relationship of a specific community health service to each health outcome is hypothesized. We focus on those community services most likely to be effective and measurable.

***ASTHMA:***

a) Effective school-based health services:
   Communication with primary care providers
   Written management plans
   Provision of monitors
   Organization and delivery of medications
   Monitoring for asthma-related school absences

b) Educational programs, such as asthma camps

c) Effective child care center-based health services Communication with primary care providers
   Family education
   Provision of monitors
   Written management plans
d) Residential control issues:
   Wood burning stove restrictions
   Control of cockroaches and other insects
   Programs emphasizing quality of public housing
e) Air quality control measures

f) Public and community efforts to increase health insurance coverage for children and youth

g) Telehealth into the homes to facilitate assessment and management

***OBESITY:***

a) Community resources enhancing physical activity: parks, playgrounds, gyms, organized programs

b) Nature of available foods: supermarkets vs. small stores

c) Public role through education or other means in controlling both the use of TV and the quality of TV for children and youth

d) Community violence than inhibits physical inactivity

e) Public programs of nutritional support: WIC, food stamps

f) School lunch programs

g) Availability of vendor foods/drinks at school

h) Community media campaigns and programs that focus on beneficial effects of diet and exercise

***RETENTION AT FIRST GRADE:***

a) Presence of child development programs

b) Role of family as compared to center-based child care

c) Early identification/intervention program

d) Home visiting programs

e) Foster care system

f) Parental literacy programs

g) Family Resource Centers

***INITIATION OF TOBACCO SMOKING:***

a) Extent of state taxation of tobacco products

b) State use of money derived from tobacco taxes

c) School policies regarding smoking on school campuses

d) Media campaigns
Control of media: Billboards, placement of advertisements Public education initiatives developed by health departments, physician organizations, health institutions

## Hypothesis 1: Community Health Services

**Statement of research question:** To what extent does state fiscal policies affect the initiation and persistence of adolescent health-related behaviors?

**Hypothesis:**

CH-1) Initiation of tobacco smoking during adolescence exhibits a high correlation with smoking rates during adulthood and, thereby, with the long-term consequences of smoking. Factors that reduce initiation of smoking during teenage years may, therefore, have great effects on modifying the disastrous consequences of tobacco smoking on the health of the public. Such factors include actions taken by state governments, most prominently through modifying the cost of cigarettes through taxation. The extent of that taxation *and* the programs put in place via use of funds derived from such taxes should be demonstrably correlated with rates of initiation of smoking.

*States with increased tobacco taxes* AND *with increased allocation of those revenues to adolescent smoking prevention and treatment programs will have the lowest rates of adolescent smoking initiation and addiction*, after controlling for individual level covariates.

CH-1a) *Social disparities in adolescent smoking rates will initially increase as the beneficial effects of state level policy on tobacco use first influence more socioeconomically advantaged youth, before diffusing to less advantaged youth.*

CH-1b) *States with increased alcohol taxes AND with increased allocation of those revenues to adolescent alcohol abuse prevention and treatment programs will have the lowest rates adolescent alcohol misuse, suggesting a generalizable model of state-level policy and adolescent health-related behaviors.*

**Rationale and significance of the hypothesis and relevance to the research question:** The relationship between public policies such as taxation and individual behaviors, such as initiation of tobacco smoking, represents a crucial example of the interface between economic policies and public health. The forms of action that are within the public sphere that have potentially significant effects on health status of individuals over the life span represent an extremely important interface between policy and health.

**Implications of hypothesis:** The hypothesis leads to direct political action as a means of improving public health. Further, the ways by which taxation-derived funds are used represents an important focus for public monitoring of the actions of state governments.

*Study design:* A large nationally representative study provides a unique opportunity to examine the joint influence of both macro-level and individual-level factors on health behaviors. Capturing the effects of state-level policy variations will require adequate sampling within *each* state to provide stable estimates of population initiation rates, including rates among important

subgroups where possible (e.g., among white vs. African-American teenagers in Rhode Island.) However, state-level sampling (to ensure representative state samples, e.g., like the PRAMS survey) does not seem warranted. Yearly sampling may be necessary to document more precisely the timing of adolescent risk behavior initiation. More realistically, a set of validated questions that assess the timing of uptake *retrospectively* will be needed. . Further, the longitudinal focus will allow mapping of changes in state policies to be correlated over time with changes in the rate of initiation of tobacco smoking.

*Sampling strategy:* This study will require a similar strategy for each state, in terms of the pattern of sampling----rural vs. urban, specific ethnic groups,
socioeconomic status, etc. Low population states may need to be oversampled to provide stable estimates of high risk populations.

*Measurement:* Rates of initiation measurement is straight forward. Given the likely survey intervals during the adolescent years, a validated set of questions for retrospective measurement of initiation is important. Measurement of the programs that tobacco taxes are used for will require
generation of a common template that can be used for each state.

***Note: The same hypothesis and subhypotheses could be appropriate for prenatal exposure to tobacco and alcohol by inserting smoking and alcohol use during pregnancy instead rather than adolescent smoking and alcohol use.***

***The longitudinal data would permit hypotheses related to the differential long term impact of prenatal exposure on subsequent child health and development based on the community services available and utilized. (Our group would be very willing to develop this hypothesis if that would seem useful.)***

## Hypothesis 2: Community Health Services

**Statement of the research question:** To what extent do health services provided in a school setting modify the natural history of chronic disease states?

**Hypothesis:**

CH-2) The nature and comprehensiveness of health services provided in settings outside traditional provider/institutional locations may exert major effects on the impact of chronic disease.

Children with asthma who attend schools with effective school-based health services, will have improved respiratory function, fewer acute health care visits and fewer asthma-related school absences, after controlling for sociodemographic and primary health care related variables.

CH-2a) School-based health services that include communication between school nurse and primary care providers, on site asthma management plans, and the capacity for direct administration of daily medicines and peak flow monitoring will be associated with the largest reductions in child morbidity.

CH-2b) Socioeconomically advantaged children with asthma will be more likely to attend schools with effective health services, while socioeconomically disadvantaged children will be more likely to demonstrate benefits from effective school health services.

CH-2c) Primary care services that articulate with school health services will lead to improved asthma outcomes.

CH-2d) Children with ADHD who attend schools with effective school-based health services, will have improved classroom behavior, fewer school disciplinary actions, and fewer school absences, suggesting a generalizable framework relating school health services and health outcomes for children with chronic conditions.

**Rationale and significance of the hypothesis and relevance to the research question:** The effect(s) of provision of health services in a school setting require ongoing inquiry as a potentially major component in relating health services to the overall health status of children and youth. The nature and extent of health services organized in the setting where children/youth function may markedly affect the natural history of the illness, use of traditional health services, rates of school absence, learning outcomes, cost of chronic disease care.

**Implications of hypothesis:** If school-based services can be demonstrated to markedly impact the natural history of a chronic disease, such as asthma, then enhancing the extent and scope of school-based health services would become an important health care strategy, and have major impact on funding, organization of services, training, etc.

*Study design:* Importance of tracking children over time in service areas where the incidence of asthma is increased, taking cognizance of the differences in rates and severity of asthma as a function of geography, ethnicity, environmental pollution, etc.

*Sampling strategy:* Oversample in areas of increased incidence of disease

*Measurement:* The relevance of school based health services for a number of important tracer conditions (asthma, ADHD, obesity, injury) offers a strong rationale for including measures of the availability and characteristics of school health services in the community. This may include pilot testing new measures. The issue of school and housing mobility are particularly important for these hypotheses.

## Hypothesis 3: Community Health Services

**Statement of research question:** To what extent do programs during early childhood modify early educational outcomes?

**Hypothesis:**

CH-3) Successful school performance and learning represents an outcome of central importance in the well-being of children and youth. Retention in grade, during early years of school, correlates directly with unsuccessful outcomes during later years of school. The capacity of children to enter the educational system effectively may be modified or enhanced if community-based programs are available for health and development promotion and early intervention of at-risk children prior to entering school. The nature and extent of *primary prevention and early identification/intervention programs* within communities becomes potentially a variable of great significance in relation to overall school outcomes.

Increased availability, quality, coordination and outreach of infancy and early child development programs will reduce the incidence of grade retention during the elementary school years, with greatest reductions evident in Grade 1.

**Rationale and significance of the hypothesis and relevance to the research question:** Programs that have demonstrably positive effects on enhancing early learning will have far-reaching implications on health outcomes, health-related behaviors, school funding, and on the nature of public responses to early child development.

**Implications of hypothesis:** If it can be demonstrated that the nature and extent of prevention and early identification/intervention programs have positive impacts on enhancing school outcome (using retention in grade 1 as a marker), support for such programs will be highlighted as an important public policy, with implications for funding, organization of community-based services, etc.

*Study design:* Need to identify workable strategy for characterizing the nature and extent of prevention and early identification/intervention programs.

*Sampling strategy:* Need to track children in areas with demonstrable differences in prevention and early identification/intervention programs, and in areas with variations in rates of early retention.

*Measurement:* Major challenge will be to work out measures of the extent and nature of early identification and intervention programs in a given community.

## Hypothesis 4: Community Health Services

**Statement of research question:**
How does community context influence the development and persistence of childhood obesity?

**Hypothesis**
CH-4) Neighborhood factors that promote healthy diets and physical activity reduce the likelihood of obesity.

CH-4a) Community violence, exposure to community violence and parental fear of community violence are all associated with decreased outdoor physical activity and increased likelihood of obesity.

CH-4b) Poor school-based nutrition, including non-nutritious school lunch and vending machines, and reduced school-based activity is associated with an increased likelihood of obesity.

**Rationale and Significance**
Obesity is a major public health problem and contributes substantially to other chronic diseases, such as diabetes and hypertension. Given obesity rates are rising dramatically over a very short period of time, understanding underlying genetic risks for obesity can only offer a partial understanding of the problem. It is critical to understand how our changing environment may be promoting increased expression of obesity risk.

**Implications**
*Study design*: A clustered design that sampled by neighborhood would facilitate the study of neighborhood level factors that may be influencing obesity rates.

*Sampling strategy*: See above. Neighborhood factors will likely only explain a relatively small fraction of the variance. However, the population attributable risk may be quite large given the number of people exposed. A large sample size will be required because of the large number of individual-level and neighborhood-level variables, and the number of SES and race/ethnicity subgroups of interest.

*Measurement:* State of the art community measurement strategies regarding community violence and recent efforts to examine school nutrition should be reviewed.

## Perinatal and Obstetric Services in the National Children's Study

### Introduction

The NCS has two formal workgroups that are developing hypotheses on the impact of events occurring during pregnancy on childhood health and development. However, since these groups largely are not focused on the impact of health services, the hypotheses in this subcommittee will focus on pregnancy-related health services research.

Background

- Prenatal care exists as a "given'" however it has to some extent been devalued in the socialization process of prenatal care. This is largely due to the fact that current studies on prenatal care do not address content and quality of prenatal care, and addresses limited outcomes (low birth weight, preterm birth). But there are multiple other outcomes that could and should be evaluated (content/quality of prenatal care, child health and development, family outcomes, and use of health services resources.
- Health care women receive before prenatal care, and over their life course may have impact on pregnancy outcomes or put another way, there is a potential for long term benefits if health services are utilized (implies access, availability, capacity, etc).
- Prenatal care results in cost savings thru potential to reduce long term disability and developmental problems.
- Prenatal care varies considerably but usually includes a series of medical services in a defined schedule of visits. It is increasingly recommended that it also include comprehensive screening or risk assessment, as well as educational, psychosocial, and nutritional services. The effectiveness of prenatal care and its individual components has not been adequately evaluated and experimental studies have not been done primarily because it is considered unethical to make random assignments of pregnant women to a control group that would not receive any prenatal care. However, every year new recommendations and practices are put forth into "routine" prenatal care without an opportunity for rigorous study.

The perinatal subcommittee of the Health Services Working Group proposes that the large cohort of the NCS would be an ideal opportunity to evaluate the efficacy and long term impact of prenatal care on the health and development of the child.

**Research Question 1:**
How do health services used by one generation affect the birth outcomes, health service utilization, and childhood health outcomes of the next generation?

**Hypothesis:**
OB-1) Health services used by one generation affects the birth outcomes, health service utilization, and childhood health outcomes of the next generation.

**Rationale and Significance:**
This hypothesis requires a longitudinal model incorporating 3 generations: the 100K cohort (G1), parents of the cohort (G0), and the kids of the cohort followed thru one year of life (G2). We anticipate 15% of this cohort will conceive during the 20 year observation period. Inherent to the global model of prenatal care is the inclusion of preconception care and the preconception period. The cohort (G1) is an ideal opportunity to evaluate the role of preconception health and health services use and its influence on birth outcomes…prospectively. Note, the best data regarding preconception characteristics will actually be available at the end of the study and will be specific to the study cohort (G1). The study as currently designed is likely to only capture data related to preconception retrospectively for G0. As proposed by the prenatal subcommittee of the Health Services Working Group, it could actually be a three generation study.

**Implications:**
The study should be of the United States and should oversample minority racial/ethnic groups and the poor (both have increased adverse pregnancy outcomes). As a three-generation study, the NCS should obtain vital statistics data (including birth certificates of siblings of the index child as well as of both parents). This study will provide a unique opportunity for longitudinal primary data collection including prenatal records, lab results and delivery records. The NCS should also link to any and all potentially useful data sets (hospital discharge data, health care utilization data, pharmacy records, etc.). Data collection should also include interview and survey information from the G0 and G1 groups

**Research Question 2:**
Is prenatal care utilization a longitudinal predictor for subsequent health services utilization?

**Hypotheses:**
OB-2a) Greater utilization of prenatal care will result in greater utilization of well-child care, sick-child care, and dental care.
OB-2b)Greater utilization of prenatal care and subsequent health services utilization will affect subsequent health behaviors, including preventive health behaviors, breastfeeding and seatbelt usage.
OB-2c) Through utilization of these services, long-term outcomes will be improved.

**Rationale and Significance:**
Women who utilize prenatal care services will have an opportunity to become familiar with a broader health care system and health services. This may lead to greater utilization of subsequent services that may be a result of greater comfort with the health care system, better understanding of the components of well-child care (e.g. knowledge of the regular schedule of immunizations), or other factors. Also, prenatal care utilization may be a part of overall continuity and/or coordination of care for the woman, the child or the entire family. Greater utilization of preventive services and earlier utilization of health services for intervention during acute illness will lead to better health outcomes as well as long-term cost savings.

**Implications:**
The study will need to evaluate whether greater utilization of subsequent services is a result of prenatal care/services or individual factors on the part of the patient (including socioeconomic status, insurance status over time, geographic variation in availability and/or composition of services, or ability of the prenatal care services to provide culturally appropriate services). Therefore, the study population should include varying socioeconomic levels, racial/ethnic groups, geographic regions, etc.

**Research Question 3:**
How does specific content of prenatal care impact clinical decisions and subsequent short- and long-term childhood outcomes?

**Hypotheses:**
OB-3a) Content of prenatal care will have a positive impact on short- and long-term childhood outcomes when that care is safe, effective, patient- and family-centered, timely, efficient, equitable and coordinated with public health, educational, and social services.

OB-3b) Content of prenatal care will have a negative impact or will have a less than maximum positive impact when that care is lacking in one of the characteristics listed above.

OB-3c) Content of prenatal care will have influence the clinical decisions that are made during pregnancy, delivery and perinatal care. These decisions will impact short- and long-term childhood outcomes.

**Rationale and Significance:**
Prenatal care exists as a "given" however it has to some extent been devalued in the socialization process of prenatal care. This is largely due to the fact that current studies on prenatal care do not address content and quality of prenatal care and address limited outcomes (low birth weight, preterm birth). However, there are multiple other outcomes that could be evaluated including content/quality of prenatal care, child health and development, family outcomes, and use of health services resources.

For example, decisions for early delivery for specific conditions (preterm labor, IUGR, preeclampsia) may have a significant impact on short- and long-term childhood outcomes. There is enough variation that we may see differences in childhood outcomes (health care utilization, health, childhood development) that we can provide exploratory data for establishing evidence for common practices (and subsequent intervention trials). Another example is the diagnosis and treatment of pregnancy related maternal conditions such as gestational diabetes, which may influence the rate of subsequent childhood obesity and development of Type 1 or Type II diabetes. A large cohort study will provide an opportunity for case/control studies evaluating whether specific interventions (e.g. antepartum testing, steroid use, psychosocial support services) impact short and long term child health outcomes.

**Implications:**
The NCS will have to determine the content and quality of prenatal services delivered to the women who participate in this study. The study should also look at prenatal services in the context of overall continuity and/or coordination of care for the pregnant women participating in the study as well as the children and the family.

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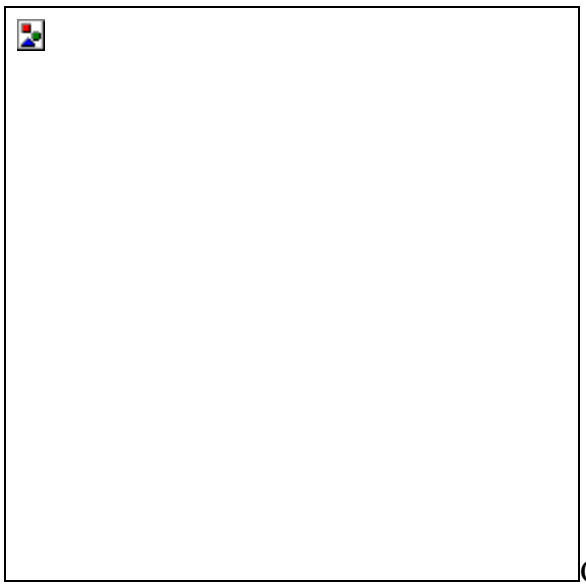

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